Case Report

Idiopathic pigmented gingival fibromatosis: A case report

Abhishek Kumar Tiwari[1], Balasundari Shreedhar[2], Attiuddin Siddiqui[3], Pratikshya Ghimire[4]

From [1]Assistant Professor, [2]Professor and Head, Department of Oral Pathology and Microbiology, Career Post Graduate Institute of Dental Sciences and Hospital, Lucknow, Uttar Pradesh, [3]Senior Resident, Department of Dentistry, Integral Institute of Medical Science and Research, Lucknow, Uttar Pradesh, [4]Lecturer, Department of Oral Pathology and Microbiology, Nepal Medical College, Kathmandu, Nepal

ABSTRACT

Gingival fibromatosis is a rare and heterogeneous group of disorders that develop as slowly progressive, local or diffuse enlargements within marginal and attached gingiva or interdental papilla. Gingival fibromatosis is a condition that can occur as an isolated disease or as a part of a syndrome or chromosomal abnormality. Here, we present the case of a 28-year-old male with pigmented gum enlargement in the maxilla and mandible. The clinical, radiographic, and histopathological features have been described in detail.

Keywords: Chromosomal abnormality, Gingival fibromatosis, Syndrome

Gingival hyperplasia is a bizarre condition causing esthetic, functional, psychological, and masticatory disturbance of the oral cavity. Familial gingival fibromatosis is a rare hereditary condition that has no definite cause. This condition may manifest as an autosomal-dominant or less commonly, an autosomal-recessive mode of inheritance, either as an isolated disorder or as part of a syndrome [1]. Idiopathic gingival fibromatosis is a rare hereditary condition that has no definite cause. It is a rare condition that is found to be associated with genetic abnormality or mutation in *Son-of-sevenless (SOS-1)* gene. Idiopathic gingival fibromatosis is a gradually progressive benign enlargement that affects the marginal gingiva, attached gingival, and interdental papilla [2]. The gingival enlargement does not occur until the eruption of the primary or permanent dentition [3]. The gingival fibromatosis has a variable clinical presentation. In some cases, it shows only minimal involvement characterized by enlargement of the tuberosity area and buccal gingiva around the mandibular molars; however, in the severe form, it can involve both maxillary and mandibular gingiva. The enlargement in severe form can be so massive that it covers the crown of both primary and permanent teeth completely.

CASE REPORT

A 28-year-old man presented with a complaint of retained roots along with swelling and pain in the upper left back region of the jaw for the past 6–8 months. The pain was dull and intermittent which often increased while lying down during nighttime. The pain gets reduced after taking painkillers. The patient also complained of difficulty in chewing due to swollen gums. His past dental history was significant with previous surgical tooth extraction for tooth number 27, 4 years back. After extraction, the patient was informed about the remaining root stumps which were left during the surgery. Now, the patient wants the removal of previously retained root stumps. The family history was non-significant.

Routine medical history and physical examination revealed a history of difficulty in hearing and some problems in the left ear for the past 8–9 years. Extraoral examination revealed a mild hypertelioric appearance. Intraoral examination revealed missing teeth 17, 18, 27, and 28, retained deciduous tooth in relation to 55, 65, and an accidental finding of gross generalized symmetric growth of gingiva in both maxillary and mandibular arches with a thick band of dark pigmentation (Fig. 1a-c). The growth was pink to pinkish-brown in color and covering almost the two-third crown portion of maxillary and mandibular teeth. On palpation, it was found to be firm and non-tender in consistency. The growth was non-fluctuant, non-compressible, and no associated bleeding was noted. Maxilla on examination revealed a generalized growth of gingiva with bilateral palatally erupted second premolars (15, 25), left rotated first premolar (24), retained deciduous (55 and 65), and spacing between the four incisors (Fig. 1c). On palpation, the mass was found to be firm and non-tender. However, intraoral examination of the mandible revealed a gross generalized growth of gingiva covering more than 2/3rd of the crown up to the incisal edge (Fig. 1b). The growth was pink to pinkish-brown in color with a grainy surface which was found to be firm and non-tender on palpation. No associated lymphadenopathy was found.

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Correspondence to: Dr. Abhishek Kumar Tiwari, E-3/62, Sector H, Aliganj, Lucknow - 226 024, Uttar Pradesh, India. E-mail: dr.abhishek0404@gmail.com

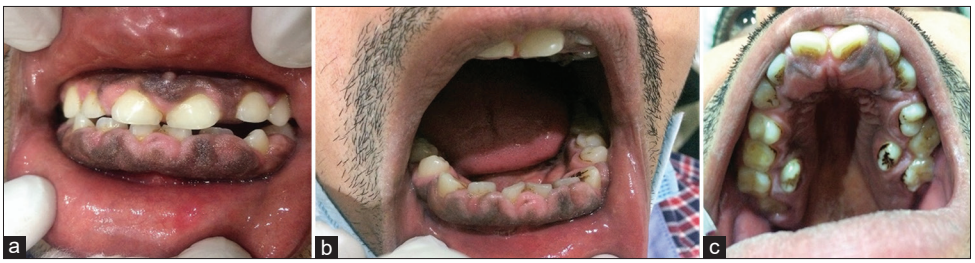

Figure 1: (a) Intraoral examination revealed gross generalized symmetric growth of gingiva in both maxillary and mandibular arches with thick band of dark pigmentation; (b) Intraoral examination of mandible revealed a gross generalized growth of gingiva covering more than 2/3rd of the crown up to the incisal edge; (c) Maxilla revealed a generalized growth of gingiva with bilateral palatally erupted 2nd premolars, left rotated 1st premolar (24) and retained deciduous 55 and 65 and spacing between the four incisors

A panoramic radiograph was advised to the patient. Multiple retained primary teeth impacted permanent teeth, and an associated radiolucency with retained root stumps in relation to 27, and carious 28 were revealed by panoramic radiograph (Fig. 2). A provisional diagnosis of idiopathic gingival fibromatosis was made after the clinical and radiological examinations.

The patient was well informed about the accidental intraoral findings and was advised for incisional biopsy along with surgical extraction of root stump of 27 and retained third molar (28). Incisional biopsy was performed along with the extraction of root stump and retained tooth. The root stump extracted was found to be hypercementosed. Soft tissue specimen was sent for routine histopathological processing and root stump for decalcification.

Histological examination revealed hyperkeratotic, hyperplastic stratified epithelium with long rete ridges with underlying densely packed, avascular connective tissue under scanner view. The bulbous increased connective tissue was relatively avascular and has densely arranged collagen-fiber bundles, numerous fibroblasts, and mild chronic inflammatory cells. The overlying epithelium is thickened and acanthotic and has elongated rete ridges. Under higher magnification, the Hematoxylin and Eosin stained sections revealed well-differentiated densely parakeratinized, hyperplastic, thick, and acanthotic stratified squamous epithelium with thin and long rete ridges (Fig. 3a). Areas of epithelium showed mild cellular atypia with nuclear hyperchromatism, and increased Nuclear/Cytoplasmic ratio. Underlying connective tissue was densely collagenous and nearly avascular with very few small blood vessels (Fig. 3b). At numerous foci, the basal layer of epithelium shows melanocytic pigmentation with numerous melanocytes with sub-epithelial chronic inflammatory cell population (Fig. 3c). On the basis of clinical, radiological, and histopathological examination, a final diagnosis of idiopathic pigmented gingival fibromatosis has been achieved.

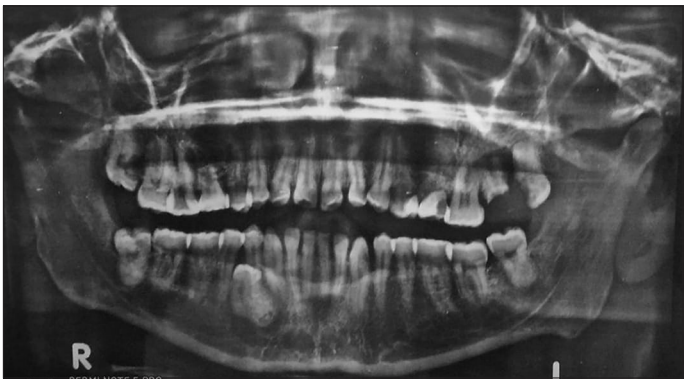

Figure 2: Multiple retained primary teeth, impacted permanent teeth and an associated radiolucency with retained root stumps in relation to 27 and carious 28 were revealed by panoramic radiograph

DISCUSSION

Gingival overgrowth varies from mild enlargement of isolated interdental papillae to segmental or uniform and marked enlargement affecting one or both of the jaws [1]. There are multiple causes of generalized gingival fibromatosis such as mouth-breathing gingivitis, drug-induced gingival overgrowth, scurvy, hereditary gingival fibromatosis, Wegener granulomatosis, acanthosis nigricans, and idiopathic variety [1]. We reported and present a case of idiopathic pigmented fibromatosis gingivae with clinical, radiological, and histopathological details of the same. General physical examination of the patient revealed no syndromic association which could contribute to fibromatosis.

The precise mechanism of idiopathic gingival fibromatosis is unknown but it appears to confine to the fibroblasts which harbor in the gingivae. The hyperplastic response does not involve the periodontal ligament and occurs peripherally to the alveolar bone within the attached gingival [2]. Gingival fibromatosis can present in two forms, a nodular form affecting the dental papillae or a symmetric uniform gingival enlargement. The enlarged tissues can partially or totally cover the crowns, cause diastemas and pseudopockets, delay tooth eruption, and malocclusion [3]. Recently, a mutation in the SOS-1 gene has been held responsible for this rare hereditary condition. Other possible causative mechanisms for hereditary gingival fibromatosis are increased proliferation and elevated production of extracellular matrix molecules, type 1 collagen, and fibronectin could contribute to the increased bulk of gingiva. Another mechanism is impairment in extracellular matrix degradation [4]. It may also develop in susceptible individuals as a side effect of systemic medications, including the anti-seizure, immunosuppressant, or calcium channel blockers [5].

The gingival fibromatosis appears as generalized nodular hyperplastic gingiva which is firm to bony hard in consistency. Usually, the hyperplastic gingiva is pale pink to red and inflamed while, in this case, enlarged gingiva is pigmented. It has a characteristic smooth to pebbled surface with little tendency to bleed. The enlargement is usually painless and may extend up to

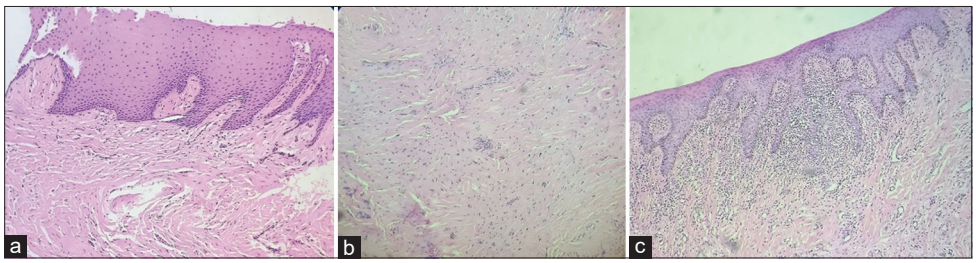

Figure 3: (a) Well-differentiated keratinized, hyperplastic, thick and acanthotic stratified squamous epithelium with thin and long rete ridges; (b) Underlying connective tissue is densely collagenous and nearly avascular with very few small blood vessels; (c) At numerous foci the basal layer of epithelium shows melanocytic pigmentation with numerous melanocytes with sub-epithelial chronic inflammatory cell population

the mucogingival junction but does not affect the alveolar mucosa or alveolar bone. The severity may vary from mild involvement of one quadrant to severe involvement of all four quadrants [6].

The precise mechanism of familial gingival fibromatosis is unknown, but it appears to be confined to the fibroblasts that harbor in the gingiva [2]. There is inconsistency in the literature as to the cellular and molecular mechanisms that lead to gingival fibromatosis. The constant increase in the tissue mass can result in delayed eruption and displacement of teeth, arch deformity, spacing, and migration of teeth [7] as, in our case, there were retained deciduous teeth as well as impacted teeth along with midline spacing and diastema. Alveolar bone is rarely affected, but the presence of pseudo-pockets and difficulty in maintaining oral hygiene may lead to some periodontal problems [8]. The extent and severity of fibromatosis in our case have covered almost all teeth in both maxillary and mandibular arch with almost covering more than 2/3rd of the crown portion in the anterior region thereby causing difficulty in mastication and speech. The differential diagnoses for the condition include drug-induced gingival enlargement, scurvy, sarcoidosis, Crohn's disease, Cowden's syndrome, and amyloidosis [8]. The treatment need varies according to the degree of severity, when the enlargement is minimum, good scaling of the teeth and home care may be essential to maintain good oral health. The relative increase in the gingival mass contemplates the need for surgical intervention owing to the functional and aesthetic compromise [9].

CONCLUSION

The presented case was of non-syndromic idiopathic pigmented gingival fibromatosis. Gingival fibromatosis is a rare disorder characterized by localized or generalized gingival enlargement. Although surgical excision is indicated as a treatment if functional impairment exists, treatment is not required in all cases of idiopathic gingival hyperplasia.

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