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Research Article

IDIOPATHIC GINGIVAL FIBROMATOSIS – A CASE SERIES AND REVIEW

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A B S T R A C T

Gingival hyperplasia is a rare condition and is of importance for cosmetic and mechanical reasons. Idiopathic gingival fibromatosis, a benign, slow-growing proliferation of the gingival tissues, is genetically heterogeneous. The enlargement is most intense during the eruption of the primary and permanent teeth, and minimal or nondetectable growth is observed in adults. The purpose of this paper is to report a case series of idiopathic gingival fibromatosis. The diagnosis was established through history and clinical examination. Surgical treatment, which included both gingivectomy and undisplaced flap with multiple teeth extraction was carried out.

INTRODUCTION

Gingival fibromatosis (GF) is a rare disease that is characterized by benign, slowly progressive, non-hemorrhagic, fibrous enlargement of maxillary and mandibular gingiva[1].The prevalence is one per 175 000 population, and men and women are equally affected[2].Clinically, the onset is consistent with the eruption of permanent dentition. At times, it is correlated to the eruption of primary dentition. It rarely presents at birth. Overgrowth can be observed varying in extent and severity. The excess gingival tissue may cover partial or whole crown, resulting in diastemas, teeth displacement, retention of primary teeth, or impacted teeth. The hyperplastic gingiva is usually normal in color, with firm consistency and heavy stippling[3].

To date, the precise pathogenesis is poorly known. Some researchers believe that the pathogenesis is confined to the fibroblasts in gingiva. A considerable number of articles support an increase of fibroblasts in GF[4]. Decreased apoptosis together with increased proliferative activity in fibroblasts could contribute to fibrotic overgrowth of gingiva. But it is still controversial if there is correlation between the amount of fibroblasts and that of collagen in all GF types, some studies show relatively few fibroblasts. Impaired balance between production and degradation of collagen may also contribute to the disease[5, 6].Research at molecular level reveals abnormal expression of some molecules related to extracellular matrix metabolism, such as transforming growth factor-β, and the abnormal expression at last leads to increased extracellular matrix deposition that contribute to pathogenesis[6].

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GF can occur as an isolated finding or as a part of a genetic syndrome, and the involvement may be generalized or localized.

Accordingly Takagi Et Al.[7], have classified GF as

1. Isolated GF;
2. Hereditary (Localized/Generalized GF);
3. Isolated idiopathic GF (Localized/Generalized);
4. GF with hypertrichosis;
5. GF with hypertrichosis and mental retardation and/or epilepsy;
6. GF with mental retardation and/or epilepsy; and
7. GF with other diseases and/or with formation of syndromes (Table 1).

Isolated GF may result from a single gene mutation, whereas syndromic forms may result from alteration in multiple genes or a single gene dosage effect. Syndromic forms of GF show dominant or recessive Mendelian transmission patterns[6]

Table 1 Syndromes associated with Gingival Enlargement

Syndrome	Genetic mode	Main characteristics	Other
Zimmerman-Laband	AD	GF, Hypoplasia of distal phalanges, Joint Mobility, Hepatosplenomegaly, Epilepsy	Hypertrichosis, Mental retardation
Jones	AD	GF, Progressive neural deafness	No
Klippel-Trenaunay	AD	GF, Hemihypertropy, Nevus flammeus, Hemagioma	Hypertension, Macrocephaly, Flat nasal bridge

Rutherfurd	X-linked	GF, Unerupted teeth, Corneal dystropy, Mental retardation	Corneal opacity, Failure of tooth eruption, Aggrssive behavior
Cross	Unknown	GF, Nanophthalmos, Microcornea, Severe mental retardation	White skin, Blond hair with yellow gray metallic sheen
Ramon	AR	GF, Hypertrichosis, Mental retardation, Epilepsy	Somatic retardation, Rheumatoid arthritis, Ocular abnormalities, Diabetes mellitus, Vascular skin lesion

AD- Autosomal Dominant , AR- Autosomal Recessive

Gingival enlargement occurs because of increased deposition of extracellular matrix (ECM), particularly interstitial collagen type I, which is one of the major components of the ECM of gingival connective tissue. Its content is determined by the balance between synthesis and degradation of matrix metalloproteinases [8]. Increased production and reduced degradation of ECM is thought to be brought about by transforming growth factor β1 (TGF-β1), a cytokine, which is an important mediator of wound healing and tissue regeneration [9]. TGF-β1 is the major mediator influencing collagen turnover and is an autocrine stimulator of hereditary gingival fibromatosis (HGF) fibroblast proliferation [10]. Fibroblasts from hereditary GF tissues produce increased amounts of TGF-β1 and β2 [11].Wright *et al*. concluded the expression of TGF-β isoforms and TGF-β receptors in hereditary and drug-induced gingival overgrowth (DIGO). They indicated differences in TGF-β isoform expression between DIGO and HGF and noted that alterations in TGF-β isoform expression by fibroblasts are implicated in the pathogenesis of both types of gingival overgrowth [12].

CASE- 1

22 year old female patient came with complaint of enlarged gingiva on left side of the both arch in dept. of Periodontology at Govt. Dental College and Hospital, Nagpur. On clinical examination it was known that patient had gingival overgrowth on left side of the both maxillary and mandibular arch. Patient gave history regarding gingival overgrowth on right side of the both maxillary and mandibular arch. Before 10 year patient had undergone surgical treatment for gingival overgrowth on right side of both the arches.

Family History-Patient's parents and siblings, grand -parents and other family member were not affected by similar condition.

Medical History-Patient's medical history was normal

On extra-oral examination slight facial puffiness was present on left side of the face which was due to gingival enlargement of both the arches on the same side. Face was well proportionate except slight depressed nasal bridge.

On intraoral Examination, the enlargement affected the attached gingiva, as well as the gingival margin and interdental papilla. The facial and lingual/palatal surfaces of the mandible and maxilla on left side were affected. The enlarged gingiva was pink, firm, and almost leathery in consistency and had a characteristic minutely pebbled surface. In posterior region of both arches teeth were almost completely covered, and the enlargement projected into the oral vestibule. The jaws appear distorted because of the bulbous enlargement of the gingiva (Fig.1).

Clinically patient's maxillary right third molar, left second and third molar and mandibular right second and third molar and left second and third molar were missing.

On radiographic examination it was found that all these teeth were impacted (Fig.2).Maxillary left first molar was grade I mobile with buccal grade II furcation involvement. Mandibular left first molar was grade III mobile. Patient had periodontal pocket with relation tomaxillary left first and second premolar and first molar and mandibular left first premolar, second premolar and first molar.

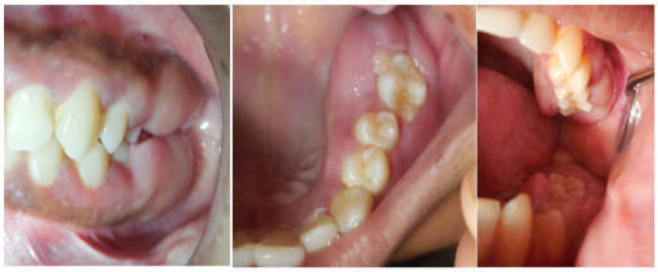

Fig 1 Clinically hyperplastic gingival tissue with extension in the vestibule and occlusal level of the teeth interfereing with the normal chewing and mastication.

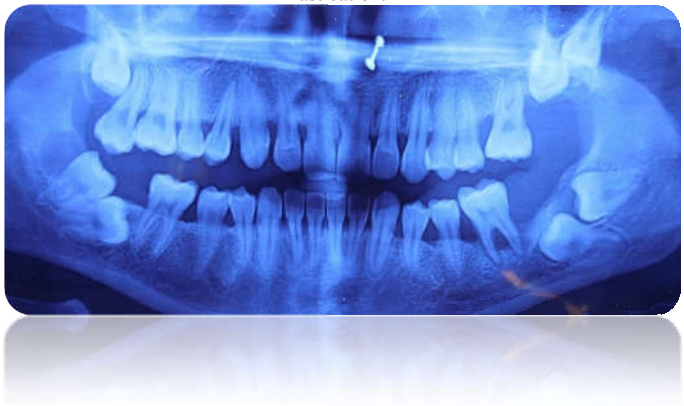

Fig 2 OPG showing multiple impacted teeth which are missing clinically

Treatment

Initially patient had undergone (before 10 years) gingival excision surgery on right side of both the arches without any tooth extraction but at the same time patient could not get treatment on the left side of the mouth. So after almost 10 years patient came again came with same complaint on left side. Patient did not have any pain but due to enlargement of tissue she had discomfort during chewing and swallowing of the food. Due to increased tissue size mechanical cleaning of the oral cavity was difficult by salivary action. Patient's oral hygiene maintenance was quite good on right side because normal gingival architecture was achieved by gingival excision.

In maxillary left posterior region undisplaced flap was performed. After proper debridement, FDBA bone graft material was filled in first molar grade II buccal furcation. Flap was respositioned and sutured with 3-0 silk suture material. Patient had periodontal pocket wrt. 34, 35 & 36. Undisplaced flap was performed in mandibular left posterior region. Second and third molar were bony impaction. Surgical removal of the

second and third molar were performed.First molar extraction was also done because it was grade III mobile. Thorough debridement was done with 34,35 region and flap was sutured with 3-0 silk suture material. Recurrence following surgical intervention is unpredictable. It is most commonly seen in children and teenagers rather than adults. In present case recurrence was not detected till 1 year of follow up period.

Recurrence is minimal or delayed if good oral hygiene is achieved by a combination of monthly examinations with professional cleaning and oral hygiene instructions. Benefits of surgical intervention are well known to improve patient's quality of life since removal of hyperplastic gingival tissue eliminates difficulties in eating and phonation, improves access for plaque control, and leads to psychological benefits due to esthetic improvement.

CASE- 2

18 year old female patient came with complaint of enlarged gingiva on left side of the both upper and lower arch in dept. of Periodontology at Govt. Dental College and Hospital, Nagpur. Familay and medical history were negative.

On extraoral examination slight facial puffiness was present on left side of the face which was due to gingival enlargement of both the arches on the same side.

On intraoral examination, the enlargement was seen affecting marginal gingiva, interdental papilla, and the attached gingiva.The facial and lingual/palatal surfaces of the mandible and maxilla on left side were affected. The enlarged gingiva was pink, firm, and leathery in consistency and had a characteristic minutely pebbled surface. In posterior region of both arches teeth were almost completely covered, and the enlargement projected into the oral vestibule. The jaws appear distorted because of the bulbous enlargement of the gingiva.Right side of both maxillary and mandibular arch gingiva were not involved, also overtained 71 and 81 were present with anterior deep bite and lingually placed 31 and 41.

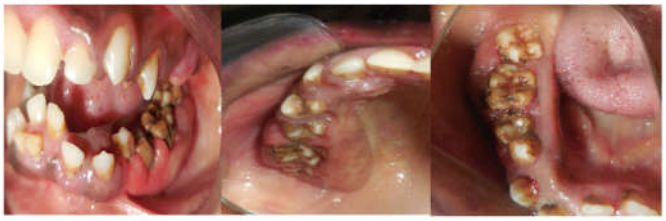

Fig. 3 clinically hyperplastic gingival tissue with extension in the vestibule and occlusal level of the teeth interfereing with the normal chewing and mastication.

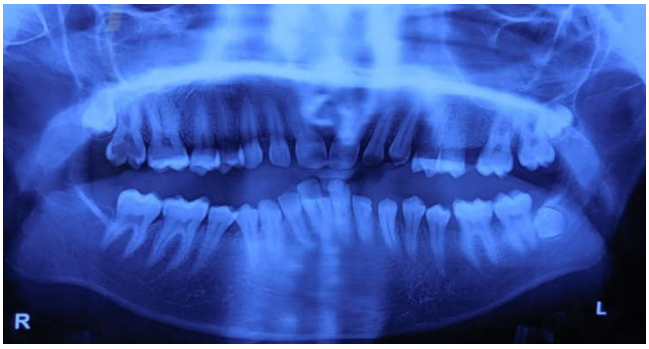

Fig. 4 OPG showing overretained 71 and 81

Treatment

Phase 1 therapy was carried out**.**After 1-month surgical phase was carried in maxillary and mandibular left posterior region where undisplaced flap was performed. After proper debridement, Flap was sutured with 3-0 silk suture material back to its original position.

CASE-3

A 14-year-old boy came with complaint of enlarged gingiva since two months in department of Periodontology at Govt. Dental College and Hospital, Nagpur.

Medical history-H/o measles & stomachache since 6 months with this reference patient went under a series of investigations with no significant findings. Suspicion of any syndrome was also ruled out after normal investigation reports.

On intraoral examination**,** the diffused gingival enlargement extending from 13 to 23 in maxillary arch and 32 to 42 in mandibular arch was observed. The enlargement was pink, firm and almost fibrous in consistency covering 1/3rd of crown surfaces (Fig.5).

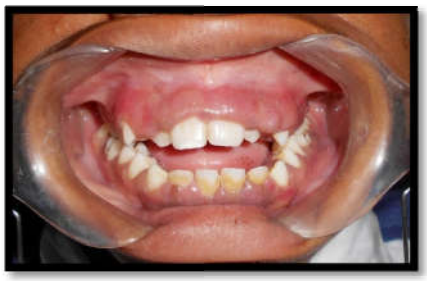

Fig 5 Clinically hyperplastic gingival tissue with extension in the vestibule and occlusal level of all teeth except 11 and 21(middle 3rd) which interfere with the normal chewing and mastication

Investigations

Based on patients medical history and clinical presentation, patient was reffered to department of paediatrics to rule out any possible syndrome and department of general medicine where he underwent a series of investigations to rule out any systemic involvement No altreration in bone involvement was observed in OPG.CBC,BT,CT levles were within normal limits.LFT, KFT values were within normal range.USG Abdomen suggested retroperitoneal lymphadenopathy suspective of tuberculous or malignant involvement but was ruled after confirmation on CECT abdomen. Finally the biopsied specimen was sent to department of oral pathology for histopathological report.

Provisional Diagnosis- Idiopathic gingival fibromatosis

Differential Diagnosis

1. Pubertal gingival enlargement,
2. Aleukemic leukemia induced gingival enlargement,
3. Tuberculous gingival enlargement.

Histopathological Report (Maxillary tissue biopsy)

- Histopathological study revealed parakeratinised stratified squamous surface epithelium with long slender,pointed fork like rete ridges.
- The underlying lesional connective tissue consists of haphazardly arranged dense collagen fibre bundles,interspersed with fibroblasts,many small & large engorged endothelial lined blood vessels,few

hemorrhagic areas & foci of moderate chronic inflammatory cellular infiltration.

Histopathological Diagnosis: Fibrous hyperplasia
(Mandibular tissue biopsy)

The underlying lesional connective tissue consists of haphazardly arranged dense collagen fibre bundles,interspersed with fibroblasts,many small & large engorged endothelial lined blood vessels,few hemorrhagic areas & foci of moderate chronic inflammatory cellular infiltration.

Few bits of lesional tissue consist of lymphoid aggregates,some of these show presence of multiple tubercles,these tubercles consist of epitheloid cells,langhans giant cells surrounded by a rim of lymphocytes.

Histopathological Diagnosis: Granulomatous lesion.

Treatment

Scaling and polishing followed by external bevel gingivectomy w.r.t maxillary anterior region. Followed by orthodontic treatment

Surgical Management-Considering the size and extent of gingival enlargement, a quadrant-wise gingivectomy was performed under local anaesthesia. An external bevel gingivectomy was done in all four quadrants. Healing was uneventful. The total masses of excised gingival tissue were sent for histopathological examination.

Final Diagnosis

On the basis of medical, family, drug history, clinical and histological findings, it was diagnosed as idiopathic gingival enlargement.

DISCUSSION

IGF may be congenital or hereditary. Though the genetic mechanism is not well understood, the majority of the reported cases have attributed the condition of fibrous enlargement of gingiva to hereditary factors. The mode of transmission is mainly autosomal dominant. The first polymorphic marker for HGF phenotype is chromosome 2p21.[13,14] Many cases are sporadic with no familial background.Gingival hyperplasia can occur after therapy with drugs like phenytoin,[15]cyclosporine, nifedipine, and nitrendipine.Gingival hyperplasia may be associated with physical development, retardation, and hypertrichosis[16]. Although gingival tissue may appear normal at birth, hyperplastic GF may become evident with the eruption of primary or permanent dentition, suggesting a trauma-induced tissue reaction during the eruption[17]. Sometimes gingival enlargement does not occur until the eruption of the permanent dentition. Further enlargement does not occur once the growth of jaw is completed[18]. It has been suggested that gingival enlargement may be due to nutritional and hormonal factors; however, these have not been completely substantiated. The constant increase in the tissue mass can result in delayed eruption and displacement of teeth, arch deformity, spacing, and migration of teeth[19].The condition is not painful until the tissue enlarges to partially cover the occlusal surface of the molars and become traumatized during mastication, which was observed in the present case. Due to massive gingival enlargement, an affected child usually develops abnormal swallowing pattern and experiences difficulty in speech and mastication. Along with these features, there may be some interference with the oral hygiene measures and normal mastication. All these will favor accumulation of materia alba and plaque, which further complicates the existing hyperplastic tissue. Maintenance of good oral hygiene is very important. It is not known if plaque control measures are effective in this condition; however, it is a good practice to maintain the plaque control following gingivectomy procedure.Since recurrence could be expected within a few months after surgery and may return to the original condition within few years, the patient may have to undergo repeated gingivectomy procedures.

In this case the final diagnosis of idiopathic gingival enlargement was made because the patient was not under any medication with no significant family history was present and the patient did not exhibit any signs of syndrome. Also, the histopathological findings were diagnosis was hyperplasia, which confirmed our diagnosis. 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 peripheral to the alveolar bone within attached gingiva. Fibromatosis gingivae may hinder tooth eruption, mastication, and oral hygiene.

Case 1-On basis of history and clinical examination it was concluded that patient was suffering from idiopathic gingival fibromatosis without any syndromic association. Possibly the pathology could have been traced back to when the patient was about 10 years of age. At the age of 10 year gingival enlargement would have occurred in all four quadrant of the mouth. Due to firm and fibrous nature of the gingival tissue second and third molar could not erupted in the oral cavity and remained impacted. Its a well know fact that gingival enlargement can cause malalignment or impaction (soft or hard tissue) of teeth [4]. In this patient 18, 27, 28, 37, 38, 47, 48 got impacted.

The finest and suggested treatment modality for idiopathic gingival fibromatosis is surgical excision by gingivectomy.

CONCLUSION

Present cases were of nonsyndromic idiopathic gingival fibromatosis, with its multidisciplinary management.The cases of idiopathic gingival enlargement cause esthetic and functional impairment which needs surgical corrections. It has a tendency to recur and patient needs to be aware of necessity of retreatment.

Conflict of Interest

There is no conflict of interest.

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