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Clinicopathological presentation of non-syndromic gingival fibromatosis and its management-report of two cases.

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Abstract

Gingival fibromatosis (GF) is a rare hereditary condition characterized by slowly progressive, non hemorrhagic, fibrous enlargement of maxillary and mandibular keratinized gingiva caused by increase in submucosal connective tissue elements which can be localized or generalized. This condition can be inflammatory, non-inflammatory or combination of both. The etiology involved could be due to poor oral hygiene, inadequate nutrition, and a systemic hormonal stimulation. Genetic cause has been implicated as its etiology with several genes mutations and sometimes associated with syndromes such as Cross syndrome, Rutherford syndrome or Ramen syndrome but isolated cases are also reported and their etiology often remains unknown.

This paper reports two very interesting case in which a4-year-old female child reported with the complaint of severe gingival enlargement with all deciduous teeth embedded resulting in difficulty in chewing, speech, and esthetics. Another patient of19 years old male patient reported with severe gingival enlargement. Primary routine investigations &detailed history led to provisional diagnosis of gingival fibromatosis in both the patients. Incisional biopsy followed by his to pathological examination confirmed the diagnosis.

Keywords: Gingiva, Gingival Fibro matosis, Gingi vectomy, Diathermy

Introduction

GF is a bizarre condition resulting inaesthetic, functional, psycho logical, and masticatory disturbance of the oral cavity. GF is an uncommon, benign, hereditary condition which is characterized by a slowly progressive, non hemorrhagic, fibrous enlargement of maxillary and mandibular keratinized gingiva.

Though many cases of GF are iatrogenic; but some are inherited while others are idiopathic[1].But still initiating factors can be due to plaque accumulation, due to poor oral hygiene, inadequate nutrition, or any systemic hormonal stimulation[2].Clinically, the hyperplastic gingival tissue is usually pale-pink, firm, has leathery consistency and presents a characteristic pebbled surface. The enlarged tissues may partially or totally cover the dental crowns, can cause diastemas, pseudo-pocketing, delay or impede tooth eruption. In severe cases it may lead to painful mastication and speech impairment. The condition may present as a nodular form characterized by the presence of multiple tumours in the interdental papillae or a more common symmetric form resulting in uniform enlargement of gingiva or a combination of both[3].It may be unilateral or bilateral, localized or generalized and can affect both maxilla and mandible. Females and males appear to be equally affected.

According to Gorlin et al in the year 1996, GF is most commonly associated with hypertrichosis, also occasionally associated with mental retardation and epilepsy. Syndromes that have been associated with Idiopathic GF are Zimmerman-Lab and syndrome (defects of bone, nail, ear and nose accompanied by splenomegaly), Murray-Paretic-Drescher syndrome (multiple dental hyaline tumours), Rutherford syndrome (corneal dystrophy), Cowden syndrome (multiple hamartomas), and Cross syndrome (hypo pigmentation with athetosis)[4]More recently, hearing loss and super numerary teeth have been reported to be associated with it.

Investigations are in evolution to establish the genetic linkage and heterogeneity associated with it[5]. 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[6]. In modern times, a mutation in the son of sevenless-1 (SOS-1) gene has been suggested as a possible cause of isolated (non-syndromic) gingival fibromatosis, but no definite linkage has been established[7].

Histopathologic ally, the bulbous increased connective tissue is relatively avascular, and has coarse and fine densely arranged collagen-fibre bundles, numerous ‘plump ’fibro blasts, and mild chronic inflammatory cells. The overlying epithelium is thickened and acanthotic, with elongated rete ridges [8].

Here We report two cases of non-syndromic cases of gingival fibromatosis along with its management.

Case reports

Case 1

A 4-year-old female patient reported with the complaint of her gingival overgrowth which in turn was creating functional and masticatory problem and led to in competent lips and poor aesthetics. Patient’s medical, dental and personal and family history was non-contributory. The patient exhibited no signs of hyper trichosis or mental retardation and had no history of epilepsy or intake of medication known to cause gingival overgrowth.

Extraoral examination revealed a convex facial profile with bimaxillary protrusion. The patient had in competent lips with flattening of nasal bridges.

Intra oral examination revealed she had pronounced generalized symmetric gingival fibromatosis covering the whole dentition both in the maxilla and mandible which was pale pinkish in colour with superimposed melanin pigmentation, fibrous in consistency (Figure 1). Bleeding on probing was absent. Gingival enlargement enclosed the major surface of the teeth present except the incisal edge of upper anterior tooth and upper molars.

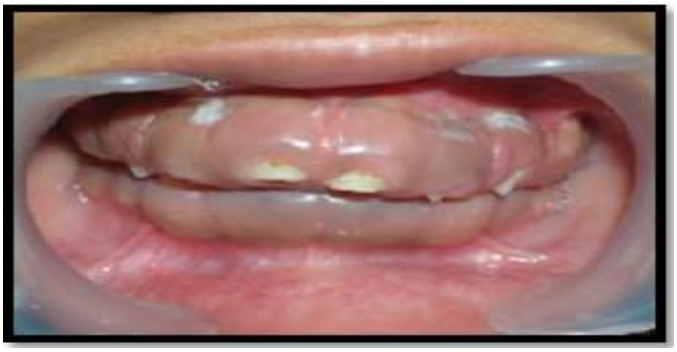

Fig 1: Preoperative frontal view of the GF

Based on the above findings, a provisional diagnosis of idiopathic generalized gingival fibromatosis was made. Radio graph ortho pantomo graphy were advised. Also, a whole-body general body examination and blood investigations were advised to eliminate any medical abnormalities.

OPG revealed (Figure 2) erupted deciduous teeth and all unerupted permanent teeth buds. No such bone loss or any significant abnormality was noted.

Fig 2: OPG showing erupted deciduous teeth and all unerupted permanent teeth buds

Surgical procedure

Considering the size and extent of gingival enlargement, gingivectomy was performed under general anaesthesia by team of oral surgeons,periodontists and anesthetists. Gingivectomy was done by diathermy and subsequently by scalpel and orbans knife to maintain the contour of the gingiva (Figures 3) . After the surgery, the site was irrigated with betadine and a Coe-pak was given for seven days. The patient was advised to take antibiotics analgesics and rinse twice daily with 0.2% Chlo rhexi dine mouthwash for two weeks.

The patient was admitted for 72 hours.Recovery was uneventful and was discharged after third post operative day.

The total masses of excised gingival tissue were sent for histopathological examination.

Figure 3:Intraoral operative view after gingivectomy

The diagnosis of gingival fibromatosis was given after histopathology examination of the excised tissues.

which revealed an acanthotic non-keratinizing stratified squamous epithelium, with elongated rete pegs. Beneath the epithelium there were dense bundles of collagenous fibrous tissue (Figure 4).

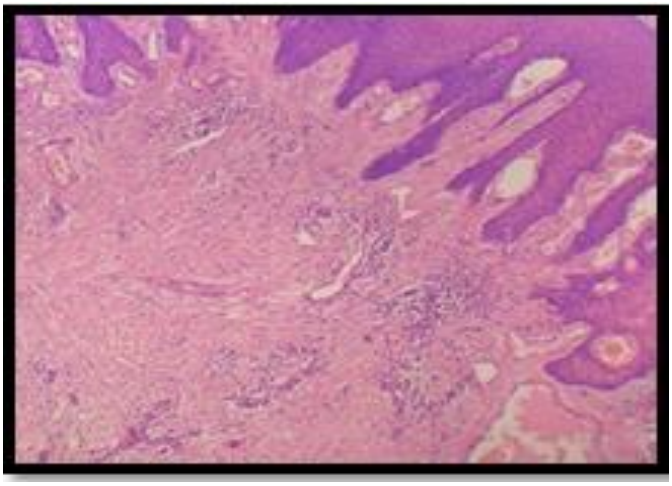

Figure 4: His to patho logical specimen showing stratified squamous epithelium with long slender rete pegs and connective tissue with dense collagen stroma

The periodontal dressing was removed after a week and healing was found to be satisfactory. The patient was then placed on a schedule of periodic recall visits for maintenance care (every three months). The oral hygiene maintenance was reinforced at every recall. No recurrence of gingival enlargement was observed one year after the surgery. (Fig. 5).

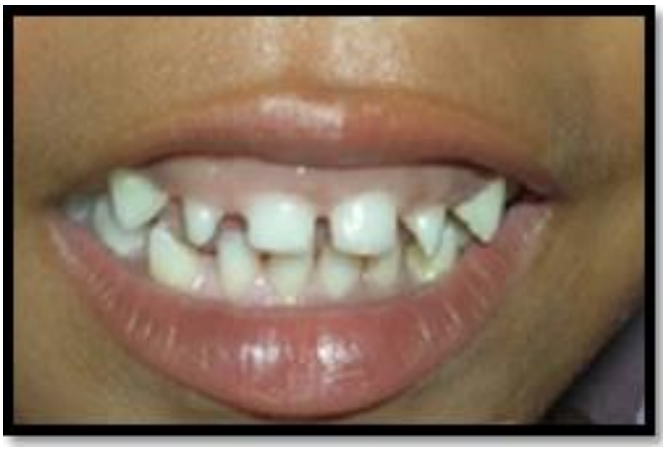

Figure 5: Postoperative view of the patient after one year

Case 2

Another patient of19 years old male patient from Murshidabad District provided with the history of bleeding gums and pain, after which swelling started to develop causing functional and masticatory difficulty. Patient's medical, dental and personal and family history was non-contributory. The patient exhibited no signs of hyper trichosis or mental retardation and had no history of epilepsy or intake of medication known to cause gingival over growth.

On intraoral examination generalized severe gingival overgrowth, involving the maxillary and mandibular arches. Enlargement involving both buccal and lingual/ palatal sides with pinkish red, fibrous inconsistency and absence of stippling. Severe diffuse enlargement involving the marginal, interdental, and attached gingiva of both arches, covering almost all the surfaces of the teeth was found (Figure 6).

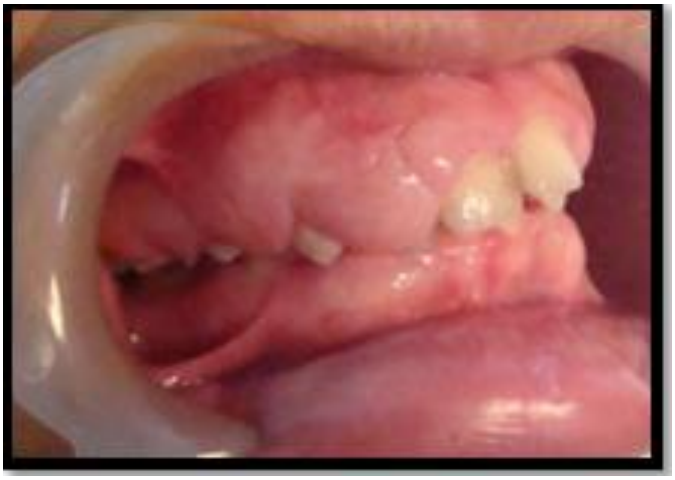

Fig 6: Preoperative photograph showing diffuse gingival enlargement

OPG was advised and was non-contributory. The blood examination results were normal and correlate d with an absence of any positive history of systemic disease. Based on all these findings, a provisional diagnosis of gingival fibromatosis was made.

Considering the size and extent of gingival enlargement, a quadrant-wise gingivectomy was performed under local anaesthesia. An internal bevel gingivectomy was done in all four quadrants. (Figure 7,8,9,10) After the surgery, the site was irrigated with betadine and a Coe-pack was given for seven days. The patient was advised to take antibiotics analgesics and rinse twice daily with 0.2% Chlorhexidine mouthwash for two weeks. Healing

was uneventful. The total masses of excised gingival tissue (Figure 11) were sent for histopathological examination (Figures 12)

Histopathological features revealed highly fibrous connective tissue, with haphazardly arranged dense collagen bundles, numerous spindle shaped fibroblasts and connective tissue that is relatively avascular. Thickened, acanthotic and hyperkeratotic stratified squamous epithelium was also present with elongated rete ridges. The histopathologic features led to the final diagnosis of gingival fibromatosis.

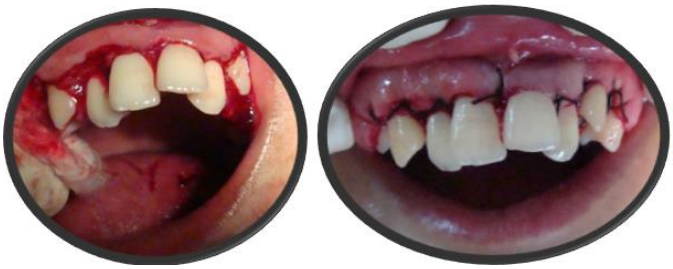

Figure 7 and 8: Intraoral operative view after gin givectomy of maxillary arch

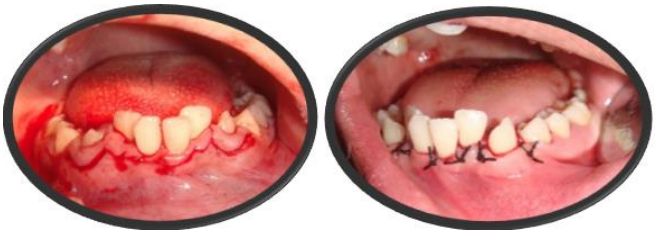

Figure 9 and 10: Intraoral operative view after gingi vectomy of mandibular arch

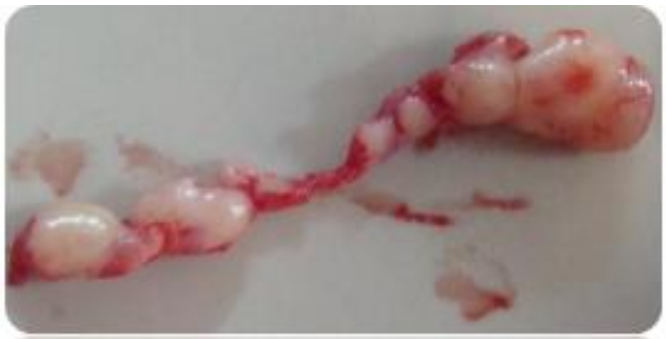

Figure 11: Excised tissue mass

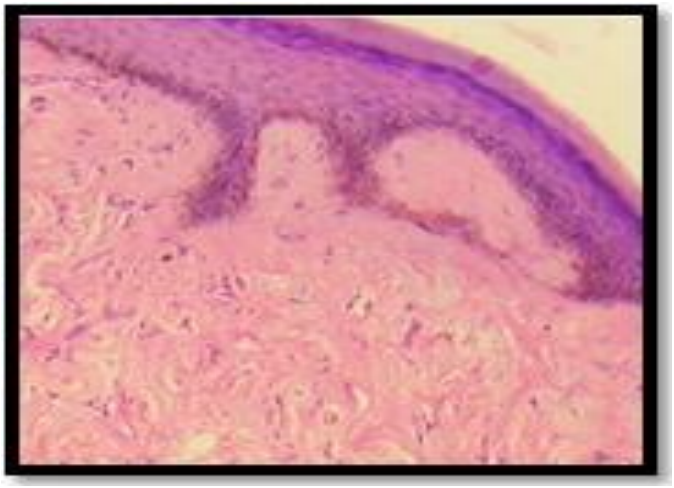

Figure 12: His to patho logical specimen showing highly fibrous connective tissue, with haphazardly arranged dense collagen bundles, numerous spindle shaped fibro blasts

The periodontal dressing was removed after a week and healing was found to be satisfactory. The patient was then placed on a schedule of periodic recall visits for maintenance care (every three months). The oral hygiene maintenance was reinforced at every recall. No recurrence of gingival enlargement was observed one year after the surgery. (Figure13).

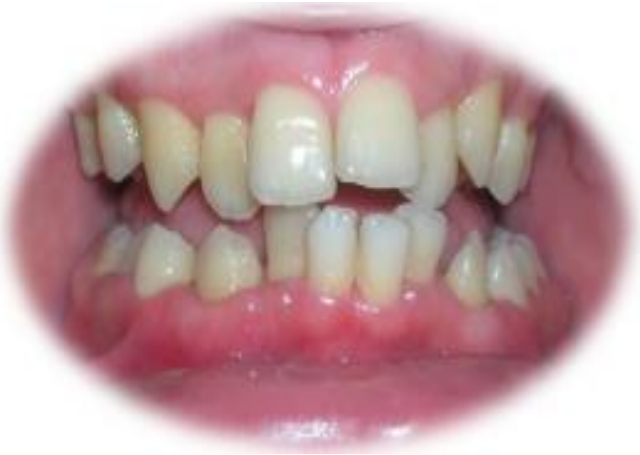

Figure 13: Postoperative view of the patient after one year

Discussion

Gingival fibromatosis is a rare, benign, non-haemorrhagic fibrous enlargement of gingival tissue varying from mild enlargement of isolated interdental

papillae to segmental or uniform or marked enlargement affecting one or both jaws [9]. It was previously called elephantiasis gingivae, hereditary gingival hyperplasia and hypertrophic gingiva [10]. It can be congenital or hereditary. Increased collagen synthesis rather than decreased levels of collagenase activity may be involved in these cases [11]. In recent studies, hearing loss and supernumerary teeth have been associated with hereditary gingival fibromatosis [12]. The condition has also been reported in association with deficiency of growth hormone caused by lack of growth hormone release factor [13]

In the present cases, the patients had no history of any systemic disease, hypertrichosis, mental retardation, epilepsy, or medication which could contribute to gingival overgrowth. General physical examination of the patient revealed no syndromic association which could contribute to gingival overgrowth. The clinical, histopathological features and systemic examination excluded the diagnosis of neo plastic enlargement, hereditary gingival fibromatosis, Wegener's granulomatosis, acanthosis nigricans [14]. The mechanism of idiopathic gingival fibromatosis is unknown, but it is seen often to confine to the fibroblasts which harbour in the gingivae. The keratinized masticatory mucosa is recognized to be developmentally unique and different tissue specific signalling pathways in this unique tissue may be responsible for the limited tissue distribution of the gingival fibromatosis phenotype[15] Severity may vary from mild involvement of one quadrant to severe involvement of all four quadrants and can even distort the appearance of jaws. Fibromatosis of the gingivae may hinder tooth eruption, mastication, and oral hygiene. In severe cases, non-eruption of the primary or permanent teeth may be the chief complaint of the patient [16]. Our first patient suffered from severe difficulty in mastication and swallowing due to gingival overgrowth which resulted in atypical swallowing pattern [17].

The gingivectomy is the surgical treatment of choice, which was first advocated for drug-induced gingival enlargement in 1941. Treatments vary according to the degree of severity of gingival enlargement. If the enlargement is mild, thorough scaling of teeth and proper home care may be sufficient to restore good oral health and appearance. However, if scaling is proved to be ineffective and the gingival overgrowth continues to affect appearance and function, surgical intervention is required. Gingival enlargement with deep pockets and severe loss of underlying alveolar bone, an internal bevel gingivectomy with open flap debridement is required [14]. Flap surgery can be carried out in areas with inadequate attached gingiva. To restore normal gingival appearance and contours gingivoplasty with blades, surgical knives, laser or electro surgery are also the treatment of choice [18].

The recurrence rate is so high that close examination was required with proper maintenance of oral hygiene. Present case has been followed for 1 year without any recurrence. Complete set of permanent teeth is the recommended time for surgery[19]. In first case eruption of complete set of permanent teeth was not present but second patient had permanent teeth, and hence surgery was considered as the best approach.

Histopathologic ally, fibromatosis shows a bulbous increase in the connective tissue which is relatively avascular and has densely arranged collagen-fibre bundles, numerous fibroblasts, and mild chronic in flammatory cells. The overlying epithelium is thickened and acanthotic and has elongated rete ridges. Our both case reports showed similar histological findings.

Conclusion

We consider these cases as an interesting case where there was excessive gingival overgrowth covering both maxilla and mandibular arches with complete presence of fibrous tissues on all the occlusal surfaces of teeth in both cases. Treatment in both the patients were done in different ways and both resulted in improvement of the aesthetic and masticatory competence as well as their periodontal condition with no recurrence till one year of follow up.

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