PERIPHERAL OSSIFYING FIBROMA – A CASE REPORT

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ABSTRACT
Peripheral ossifying fibroma is a common, benign, reactive lesion of connective tissues of jaws grouped under fibroosseous lesions, caused by prolonged irritations to gingiva by various factors. We report a case of 36 years old male patient with recurrent slow growing gingival mass in mandibular molar region. A brief highlight of literature review on occurrence rates, clinical presentations and radiologic features are discussed in this article with an overview on etiopathogenesis and management of such lesions.

KEYWORDS: Ossifying fibroma, Fibro osseous lesion, Irritation Fibroma.

INTRODUCTION
Peripheral Ossifying Fibroma (WHO 2005) (Peripheral – “surrounding” ossifying – “calcium deposition” latin - fibra, “fiber” + Greek- oma, “tumor”) also called “alveolar exocytosis “is usually a benign localised reactive lesion of the connective tissues grouped under fibro osseous lesions of jaws (WHO 2000). Peripheral fibroma with calcification, calcifying fibroblastic granuloma, Ossifying fibroid epulis, Peripheral cementossifying fibroma, Calcifying fibroma are the parallel terms used to describe this lesion. The local lesions are primarily attributed to various irritations from trauma by sharp tooth, biofilm, calculus, restorations, orthodontic appliances and hormonal influences to gingiva. Sporadic cases of such lesions are also reported in familial forms. Here, we present a case report of peripheral
ossifying fibroma of right mandibular molar region in 36 years old male patient, uncommonly presenting in the third decades of life.

Case report
A 36 years old male patient reported to outpatient department of oral medicine and radiology with the chief complaint of pain and swelling in the right lower back teeth region since a year associated with halitosis. Patient noticed a painless mass, slow growing which eventually got ulcerated to cause pain. Patient gave a history of similar swelling with pain before one and half year with recurrent bleeding. He underwent scaling and root planning followed by surgical removal of the swelling. The swelling was excised two weeks after the periodontal therapy and histopathologic examination revealed it to be pyogenic granuloma. Patient was followed up for one year and his oral hygiene was maintained. He gave no history of debilitating systemic diseases, allergy and autoimmune diseases. On intra oral examination a nodular mass of 3 cms * 2 cms was noted in the interdental region of 46 47 with superficial ulceration and centrifugal presentation. On palpation the lesion was firm, non-tender and slight tenderness with blanching was noted on compression. Radiologic examination with IOPA showed only loss of lamina dura, widening of PDL space and horizontal loss of bone with no evident bony changes.

Fig. 1: Nodular mass evident in relation to 46 and 47 interdentally.

Fig. 2: Nodular mass showing centrifugal presentation.
Fig. 3: Intra oral periapical radiograph showing alveolar crestal bone loss.

On occlusal radiograph, no evident change with respect to buccal and lingual cortical plates. Presence of thick bands of calculus covering one third tooth surfaces on both palatal and buccal aspect of all the posterior teeth was evident. Routine blood investigations including complete blood count were advised and were found to be within the normal limits. The lipid profile and levels of thyroid in the blood were at normal range; tests for hepatitis A, B and C were negative. Panoramic radiographs showed normal findings, with a moderate amount of horizontal bone loss in relation to upper anterior teeth. Patient was advised to restore oral hygiene by scaling and root planning procedure. After two weeks, excisional biopsy was done and the soft tissue was sent for histopathologic investigations.

Fig. 4: Cementum and bone like ossification in fibrocellular stroma.
DISCUSSION

Peripheral ossifying fibroma is one of the several common reactive inflammatory hyperplastic lesions of gingiva.\(^1\) Among plethora of lesions occurring in the gingiva which includes pyogenic granuloma, peripheral giant cell granuloma, peripheral fibroma and giant cell fibroma, peripheral ossifying fibroma accounts for about 2-9% of the gingival overgrowths.\(^2\) It is the third most common reactive lesion of gingiva after pyogenic granuloma and peripheral giant cell granuloma.\(^3\) Peripheral ossifying fibroma is predominant in white population (71%). The female to male ratio reported in the literature varies from 1.22:1\(^3\) and 1.7:1 to 4.3:1.2.\(^4\) POF occurs as a slow growing, painless masses unifocal or multifocal in nature, nodular or lobular, commonly occurring in maxilla (60%) predominantly in incisor region (45%) and cuspids with a greater rate of recurrence of about 16%.\(^2,3\) These lesions may arise as a result of irritants such as trauma, microorganisms, plaque, calculus, restorations, and dental appliances.\(^2\)

POF as a separate entity was first described by Shepherd et al as alveolar exocytosis in 1844. Menzel et al in 1872 reported case of ossifying fibroma with cementum like substances and named it as “Cemento ossifying fibroma”. In 1971, WHO grouped various lesions containing cementum like substance as fibrous dysplasia, cementifying fibroma, ossifying fibroma and cemento ossifying fibroma. Eversol and Robin et al coined the term “Peripheral Ossifying Fibroma” in 1972. In 1982, Gardener suggested the use of the term peripheral ossifying fibroma. Waldron and Kramer et al, classified the fibro osseous lesions of the jaws in which they described fibrous dysplasia, cementoossifying fibroma and ossifying fibroma. In 1992, the two lesions cementifying fibroma and ossifying fibroma which were described as separate
entities are given a common term cementossifying fibroma. Later, with the evidence of lamellar/woven bone deposition, dystropic calcifications and cementum like substances (cementoid) these lesions were named as “peripheral ossifying fibroma”. The term cementossifying fibroma, was scientifically inaccurate attributed to the presence of such cementum like substances in other bones such as tibia, femur and skull bones where they are named as peripheral ossifying fibroma.

There are several terms used to describe the nomenclature of this lesion. Peripheral cementossifying fibroma, peripheral fibroma with calcification, ossifying fibro epithelial polyp, calcifying fibroblastic granuloma, ossifying fibroid epulis, peripheral fibroma with cementogenesis, calcifying or ossifying fibroma epulis are the various terms used to describe the lesion earlier in the literature. WHO in 2005 revised all the calcifying fibrous lesions and grouped such lesions under an umbrella term of peripheral ossifying fibroma. They are considered non neoplastic due to slow growing and non-destructive nature. In 2017, these lesions were grouped under odontogenic lesions. The increased number of names used in naming of fibroblastic gingival lesions indicates that there was always some controversy surrounding in grouping of these lesions. When bone predominates, the lesions are termed as “Ossifying”. When there was presence of curvilinear trabeculae or spheroidal calcification then the term “Cementifying” was used. When bone and cementum like tissues are observed, the term was combined as “Cemento ossifying fiboma”. Irrespective of the nomenclature, these variant lesions are indistinguishable radiographically.

There are two basic theories involved in the pathogenesis of ossifying fibroma which includes formation from pleuripotent stem cells in the PDL ligament and chronic inflammation from pyogenic granuloma. There are also reported sporadic cases of multifocal fibromas attributed to mutations in various genes such as hprt2 gene of parafibromin proteins. Endo et al[5] in his study, compared three groups of lesions namely cementifying fibroma, ossifying fibroma and fibrous dysplasia. The results in immunohistochemical staining showed increased uptake of keratan 4 sulfate in cementifying fibroma whereas, there was increased uptake of chondroitin 4 sulfate in ossifying fibroma and fibrous dysplasia attributed to bony tissues in these lesions. All these terms are put under umbrella term of “peripheral ossifying fibroma” in 2007 by WHO irrespective of the content elaborated by the lesion.

The etiology attributing to ossifying fibroma is unclear, although there are reports of origin from periodontal ligament mesenchymal blast cells. This condition arises as a response to
long-lasting chronic stimuli. This can occur when gingival tissue reacts in response to local irritants such as biofilm, subgingival calculus, misplaced teeth, over-contoured restorations, inadequate dentures, debris from root, foreign bodies into the gingival sulcus, and injuries caused by orthodontic treatments.\cite{2}

Although the underlying etiology is uncertain, there are several reports of past trauma in the areas of the lesion. The same was evidenced in our case report. This point of trauma as a possible triggering factor in some presentations of the lesion, explains it to be a connective tissue reaction rather than a genuine neoplasm. They are also associated with systemic conditions such as in parathyroid tumors, multifocal gigantiform lesions.

POF radiographically appear as an unilocular lesion, the radio opacity of the lesion is attributed to the degree of calcification. POF can cause resorption of the alveolar crest and separation of adjacent teeth with pathologic migration (Poon et al.). Three radiologic features attributed to stages of development in ossifying fibroma namely the early, intermediate and mixed stages.

The etiopathogenesis of peripheral ossifying fibroma is uncertain in that the origin is attributed to periodontal ligament due to three reasons. The close proximity of the gingiva to periodontal ligament, recurrent lesions in the interdental gingiva and presence of oxytalan fibres in the matrix of these lesions.\cite{6} Excessive proliferation of mature fibrous connective tissue is a response to gingival injury, gingival irritation, subgingival calculus or a foreign body in the gingival sulcus.\cite{7} Chronic irritation to gingiva causes metaplasia of the bone resulting in dystrophic calcification.\cite{7}

The clinical feature of this lesion is often described as a pedunculated or sessile nodular mass\cite{5} and sessile-based is the most frequent presentation\cite{7} Regarding location, 50% of the lesions affect the region of incisors and more than half of it is located in upper arch\cite{8} (60%). Clinically, POFs are sessile or pedunculated, ulcerated or exhibiting a color similar to the surrounding gingiva.\cite{2,9,10,11,12,13} Kfir et al. reported that the size of the POF is usually smaller than 1.5 cm in diameter.\cite{14} However, a case of giant POF measuring 9 cm has been reported in the literature. Multicentric POF occurring in the oral and maxillofacial region was observed in genetic associated conditions such as nevoid basal-cell carcinoma syndrome, multiple endocrine neoplasia-Type II, neurofibromatosis, and Gardner syndrome.\cite{11}
Mesquita et al. reported a high proliferative activity of the lesion, which eventually influences the treatment modalities.\cite{15}

Due to the high recurrence rates, the treatment of POF comprehends total excision of the lesion including periosteum and PDL at the base of the lesion, after elimination of local etiologic factors such as plaque, calculus, ill-fitting dentures, and overhanging restorations.\cite{2,9,10,11,12}

The definitive diagnosis of POF is made usually by the histopathologic evaluation of biopsy specimen. The histologic spectrum of POF is wide and has been described in detail by Buchner and Hansen.\cite{20}

There are some interesting features attributed to radiographic appearances and crystalline structures of peripheral ossifying fibromas. Upon radiographic evaluation, calcifications were observed in shadow of soft tissue and rarely, there was presence of diffuse opaque calcifications with associated bone destruction. Regarding radiographic appearance of tooth migration, it is present only in 5% of the cases, thus constituting a very rare finding, as well as radicular resorption.\cite{21,10,22,23} The crystalline structure of the lesion progresses with the ageing of the lesion that causes evident radiographic characteristics.\cite{6} The arrangement of crystals in ossifying fibroma are diffuse than in bony apatite crystals showing diffuse radiographic appearance or no evident radiographic features.\cite{24} The cementicles present are not from the cementum and they represent dysmorphic product of the tumor analogous to keratin pearls in squamous cell carcinoma.\cite{7} Radiographically, POF may not show significant changes in certain cases. In some cases, varying radiodensity within the lesion was seen depending on the degree of mineralization. Superficial bone loss, cupping defect, and focal areas calcification have been rarely reported like in the present case. Additional investigations such as computed tomography and magnetic resonance imaging are also helpful in larger lesions.\cite{2,3} Presence of Saucerization was also evident in such cases of ossifying fibroma. Pathologic migration of adjacent teeth was most common. No evident root resorption is seen in patients. CT and MRI images are taken for larger lesions and the lesions which are evidently invasive. CT shows expansible mass with intact cortical plates and margins distinguishable from that of adjacent cortical bone. Soft tissue opacifications are evident within the lesion with presence of ossicles.
In haematological investigations no significant alterations are reported. Increased macrophages are characteristic feature in tissue specimens of such reactive inflammatory lesion. Macrophage count in case of peripheral giant-cell granuloma was significantly higher than all other reactive lesion of the oral cavity. Presence of genetic mutations in CD68 on karyotyping was reported in these cases. There are various stains used to describe the lesion such as Masson trichrome stains and immunohistochemistry can be done to identify oxtalan fibres to differentiate it from granuloma. Type 1 collagen immunoreactivity of ossifying fibroma was significantly higher compared to PGCG counterparts. Type 3 collagen immunoreactivity of PGCG counterparts in intermediate lesions was significantly higher compared to ossifying fibromas. Collagen type 1 forms the main component of bone and tendons showing great resistance to the tension forces. On the other hand, collagen type 3 is organized ideally as a loose mesh.

The differential diagnosis of these lesions include pyogenic granuloma, peripheral giant cell granuloma, epulis and gingival fibroma. Incisional biopsy is advocated for larger lesions of multifocal nature.

Histologically, the POF appears as nonencapsulated mass of cellular fibroblastic connective tissue primarily of mesenchymal origin, surrounded by stratified squamous epithelium, which showed ulcerated regions in almost 23%–66% of cases. Most ulcerated lesions occur in patients in the second decade. POFs contain areas of fibrous connective tissue, endothelial proliferation and mineralization. Endothelial proliferation can be profuse in the areas of ulceration, which can mislead clinical diagnosis owing to their appearance as a pyogenic granuloma.

The features observed during microscopic evaluation include benign fibrous connective tissue with varying content of fibroblasts, myofibroblasts, collagen and sparse to profuse endothelial proliferation having mineralized material which may represent mature, lamellar or woven osteoid, cementum like material, or dystrophic calcifications.

Neville et al suggested the removal of the lesion down to the adjacent periosteum and the adjacent teeth be scaled to remove any remaining irritants. Any other identifiable irritant like an ill-fitting dental appliance or rough restorations if present, should be removed. If the lesion is present in esthetic zone, reconstructive surgery should be performed to repair the defect. Different surgical techniques like coronally positioned flap, lateral sliding flap,
subepithelial connective tissue graft may be used to manage this defect and minimize patient esthetic concerns.[28]

There are cases in the literature that used the diode laser to enucleate the lesion, stating it as more advantageous because it provides a better visualization in the surgical field, faster healing, less postoperative pain, and short recovery, as well as a more satisfactory patient adherence.[29] However, high cost is its main disadvantage.[30] A Close postoperative follow-up is essential because of increased growth potential of incompletely excised lesions and the 8%–20% recurrence rate.[31] The rate of recurrence has been reported at 8.9%,[20] 9%,[32] 14%,[33] 16%[22] and 20%.[21] by various authors respectively. Therefore, regular follow-up is required. From a survey on 27 POF patients, 30.4% (8.2 patients) relapsed.[27]

Malignant transformation has been reported only for peripheral ossifying fibromas of multicentric and giant forms. It has been reported in 2 percent of overall gigantiform cases. The Recurrence rate of 16% has been reported in the literature in the 5 years follow up cases. The recurrence rate of peripheral ossifying fibroma has been considered high for reactive lesions. Recurrence in the present case can be due to previous incomplete surgical removal of the lesion.

CONCLUSION

Summing up, Peripheral cemento-ossifying fibroma is a non-neoplastic enlargement of the gingiva that is classified as a reactive hyperplastic inflammatory lesion. In majority of cases, ossifying fibromas should not be misdiagnosed as other reactive lesions arising from the gingiva. Therefore, careful clinical and histopathological examination is essential for an accurate diagnosis, proper management and adequate follow up of such cases.

REFERENCES


