Peripheral Ossifying Fibroma: A Case Report and Review

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ABSTRACT

Various types of localized reactive hyperplasia occur in the gingiva, as a result of chronic irritation from trauma, microorganisms, plaque, calculus, restorations and dental appliances. Peripheral ossifying fibroma (POF) is a non-neoplastic enlargement of the gingiva that is reactive in nature. Though the etiopathogenesis of is uncertain, an origin from cells of the periodontal ligament has been suggested. They are generally below 2 cm in diameter. POF tends to occur in the 2nd and 3rd decades of life. Almost two thirds of all cases occur in females, with a predilection for the anterior maxilla. The POF lesion does not require imaging beyond radiographs. Treatment consists of conservative surgical excision and scaling of adjacent teeth. Herewith, we present a case of POF that occurred in a 35-year-old female in the posterior maxillary gingiva, which is relatively rare.

Keywords: Peripheral ossifying fibroma, Fibrous epulis with calcification/ossification, Peripheral fibroma with ossification, Reactive hyperplasia, Radiopaque foci.


Source of support: Nil

Conflict of interest: None declared

INTRODUCTION

Many types of localized reactive lesions may occur on the gingiva, including focal fibrous hyperplasia, pyogenic granuloma, peripheral giant cell granuloma and peripheral ossifying fibroma (POF).1-3

These lesions may arise as a result of such irritants as trauma, microorganisms, plaque, calculus, restorations and dental appliances.2,3

POF is a non-neoplastic enlargement of the gingiva that is thought to be reactive in nature. Considerable confusion has existed over the nomenclature of this lesion, and several terms have been used to describe its variable histopathologic features, one of which is peripheral cemento-ossifying fibroma due to the presence of cementum-like calcifications. The pathogenesis of this lesion is uncertain and it is thought to arise from the peristeal and periodontal membrane.4

Herewith, we report a case of POF occurring in a 35-year-old female along with the review of literature.

CASE REPORT

A 35 years old female reported to the department of Oral Medicine and Radiology in Tamil Nadu Government Dental College and Hospital with the chief complaint of small growth in the gums in relation to right upper back tooth region, with duration of 5 months (Fig. 1).

History revealed that the growth was noticed 5 months back as a small one and progressed slowly to the present size. There was no history of pain or bleeding. Past medical and past surgical history were noncontributory. Past dental history revealed that the patient had undergone extraction of one right upper molar 1 year back. Personal history was not relevant.

On intraoral examination there was a well circumscribed, sessile growth measuring 2 × 0.7 cm in size, located in right upper buccal gingival 16 and 17, extending anteriorly from the middle 1/3rd of buccal surface of 16, posteriorly up to the middle 1/3rd of buccal surface of 17, superiorly up to the buccal vestibule and inferiorly up to the level of occlusal surface of 16, 17. The mucosa over the growth was smooth and normal in color. The surface appeared lobulated. On palpation it was hard in consistency, nontender and fixed. Grade III mobility was elicited in 16 and 17. The patient had poor oral hygiene with OHI-S score of three with significant amount of supragingival/subgingival plaque around all molars, specifically more at the site of the lesion (Fig. 2). Correlating the history and clinical findings the case was provisionally diagnosed as POF with a differential diagnosis of fibrous epulis.

Intraoral periapical radiograph showed missing 15 and 18 with horizontal bone loss in between 16 and 17 up to apical third regions. There was loss of lamina dura and widening of periodontal ligament space in relation to the roots of 16 and 17 (Fig. 3). The maxillary occlusal radiograph showed a soft tissue radiopaque shadow with

Fig. 1: Frontal view of the patient
irregular radiopaque areas arising in continuity with the buccal cortical bone in relation to 16, 17 interspersed throughout the soft tissue radiopaque shadow. The internal radiopacity had density similar to bone (Figs 4 and 5). Hence, the case was radiographically suggestive of POF. Routine blood and urine investigations were carried out and the values were within normal limits. The patient was subjected to scaling followed by excisional biopsy of the lesion.

The histopathological examination showed hyperplastic stratified squamous epithelium. The underlying fibrous connective tissue showed areas of ossification and inflammatory cells, confirming the diagnosis of POF (Fig. 6).

DISCUSSION

Intraoral ossifying fibromas have been described in the literature since the late 1940s. Many names have been given to similar lesions, such as epulis, peripheral fibroma with calcification, POF, calcifying fibroblastic granuloma, peripheral cementifying fibroma, peripheral fibroma with cementogenesis and peripheral cemento-ossifying fibroma. The sheer number of names used for fibroblastic gingival lesions indicates that there is much controversy surrounding the classification of these lesions. Considerable confusion has prevailed in the nomenclature of POF with various synonyms being used, such as peripheral cementifying fibroma, ossifying fibroepithelial polyp, peripheral fibroma with osteogenesis, peripheral fibroma with cementogenesis, peripheral fibroma with calcification, calcifying or ossifying fibrous epulis and calcifying fibroblastic granuloma. Lesions involving the gingival soft tissues are rare compared to the lesions appearing within bone.

Though the etiopathogenesis of POF is uncertain, an origin from cells of the periodontal ligament has been suggested. The following reasons are attributed for considering the periodontal ligament origin for POF (1) exclusive occurrence of the POF in the gingiva (interdental papilla), (2) the proximity of the gingiva to the periodontal ligament, and (3) the presence of oxtalan fibers within the mineralized matrix of some lesions. 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. Chronic irritation of the periosteal and periodontal membrane causes metaplasia of the connective tissue and resultant initiation of formation of bone or dystrophic calcification. It has been suggested that the lesion may be caused by fibrosis of the granulation tissue.

Peripheral ossifying fibroma tends to occur in the 2nd and 3rd decades of life, with peak prevalence between the ages of 10 and 19. Almost two, thirds of all cases occur in females, with a predilection for the anterior maxilla. By most reports, the majority of the lesions occur in the second decade, with a declining incidence in later years.
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There are two reported cases of POF present at birth, presenting clinically as congenital epulis.\textsuperscript{13,14} In a 2001 study, Cuisia and Brannon\textsuperscript{11} reported that only 134 out of 657 diagnosed POFs (20\%) were in the pediatric population (0-19 years), with 8\% in the first decade. In a retrospective study of 431 cases in the Chinese population by Zhang and others,\textsuperscript{15} the mean age of incidence of POF was found to be 44 years, which is contradictory to previously published literature. POF appears to be more common among white people than black\textsuperscript{16} and slightly less common among those of Hispanic origin.\textsuperscript{17}

POF may present as a pedunculated nodule, or it may have a broad attachment base.\textsuperscript{1,16,18}

These lesions can be red to pink with areas of ulceration, and their surface may be smooth or irregular. Although they are generally less than 2 cm in diameter,\textsuperscript{11} the size can vary; reports range from 0.2 to 3 cm diameter.\textsuperscript{10} Cases of tooth migration and bone destruction have been reported, but these are not common.\textsuperscript{10} The size of the POF ranges from 0.4 to 4 cm according to a different study.\textsuperscript{1} At its greatest dimension, the average lesion measures approximately 1 cm.\textsuperscript{1}

POF is thought to be either reactive or neoplastic in nature.\textsuperscript{19} It is a fairly common lesion, comprising nearly 3\% of oral lesions biopsied in one study,\textsuperscript{1} approximately 1 to 2\% in other studies.\textsuperscript{10,12,16} In 1993, Das and Das\textsuperscript{17} obtained similar results, with 1.6\% POFs among 2,370 intraoral biopsies.

Ossifying fibromas elaborate bone, cementum and spheroidal calcifications, which has given rise to various terms for these benign fibro-osseous neoplasms. When bone predominates, the term ‘ossifying’ is applied and the term ‘cementifying’ is assigned when curvilinear trabeculae or spheroidal calcifications are encountered.\textsuperscript{20} When bone and cementum like tissues are observed the lesions have been referred to as cemento-ossifying fibromas.\textsuperscript{20} Cementifying fibromas may be clinically and radiographically impossible to separate from ossifying fibromas.\textsuperscript{19}

Mesquita RA found higher numbers of argyrophilic nucleolar organizer regions (agNORs) and proliferating cell nuclear antigen (PCNA)-positive cells in ossifying fibroma than in POG indicating higher proliferative activity in ossifying fibroma.\textsuperscript{21}

X-ray diffraction analysis indicated that the mineral phase of both central and peripheral tissues consists of apatite crystals and that the crystallinity of these apatites is lower than that of bone apatite. Also, it was suggested that the crystallinity of the apatites might improve progressively with the development of the lesion, possibly to the same degree as that of bone apatite.\textsuperscript{22}

POFs are believed to arise from gingival fibers of the periodontal ligament as hyperplastic growth of tissue that is unique to the gingival mucosa.\textsuperscript{1-3,23} This hypothesis is based on the fact that POFs arise exclusively on the gingiva, the subsequent proximity of the gingiva to the periodontal ligament and the inverse correlation between age distribution of patients presenting with POF and the number of missing teeth with associated periodontal ligament.\textsuperscript{11,16,23}

In a study of 134 pediatric patients with POF,\textsuperscript{16} in only two cases was POF intimately associated with primary teeth, bringing into question the reactivity of the lesion. The exfoliation of primary teeth and eruption of their successors should result in an increased incidence of periodontal ligament-associated reactive lesions.\textsuperscript{2,16}

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Fig. 5: Cropped occlusal radiograph image shows a soft tissue radiopaque shadow on the buccal aspect of 16, 17 with irregular radiopaque foci interspersed throughout the lesion

Fig. 6: Photomicrograph showing hyperplastic stratified squamous epithelium with underlying connective tissue with areas of calcification and inflammatory cells
Hormonal influences is suspected to play a role, given the higher incidence of POF among females, increasing occurrence in the second decade and declining incidence after the third decade.10 In our case, the patient was in her fourth decade.

In an isolated case of multicentric POF, Kumar and others6 noted the presence of a lesion at an edentulous site in a 49-year-old woman, which once again raises questions regarding the pathogenesis of this type of lesion.

Histologically, the POF appears to be a nonencapsulated mass of cellular fibroblastic connective tissue3 of mesenchymal origin, covered with stratified squamous epithelium, which is ulcerated in 23 to 66% of cases.1,11 The histopathological findings of the case reported herewith is similar to the above findings. Most ulcerated lesions occur in patients in the second decade.2

Radiographic features of the POF vary. Radiopaque foci of calcifications have been reported to be scattered in the central area of the lesion, but not all lesions demonstrate radiographic calcifications.1 Underlying bone involvement is usually not visible on a radiograph. In rare instances, superficial erosion of bone is noted.1 The POF lesion is generally small and does not require imaging beyond radiographs.3,24 In our case, there was typical internal radiopaque foci which were irregular extending from the buccal cortical plate region interspersed throughout the soft tissue radiopacity.

A confirmatory diagnosis of POF is made by histopathologic evaluation of biopsy specimens. The following features are usually observed during microscopic examination: (1) intact or ulcerated stratified squamous surface epithelium; (2) benign fibrous connective tissue with varying number of fibroblasts; (3) sparse to profuse endothelial proliferation; (4) mineralized material consisting of mature, lamellar or woven osteoid, cementum-like material or dystrophic calcifications; and (5) acute or chronic inflammatory cells in lesions.1,3

Eversole and Rovin2 stated that, with the similar sex and site predilection of pyogenic granuloma, peripheral giant cell granuloma and POF, as well as similar clinical and histologic features, these lesions may simply be varied histologic responses to irritation. Gardner3 stated that POF cellular connective tissue is so characteristic that a histologic diagnosis can be made with confidence, regardless of the presence or absence of calcification.2,25

It has been suggested that the POF represents a separate clinical entity rather than a transitional form of pyogenic granuloma, peripheral giant cell granuloma (PGCG) or irritation fibroma.1

Buchner and Hansen11 hypothesized that early POF presents as ulcerated nodules with little calcification, allowing easy misdiagnosis as a pyogenic granuloma.11

When presented clinically with a gingival lesion, it is important to establish a differential diagnosis. In this case, the clinical features led to a differential diagnosis of irritation fibroma, pyogenic granuloma or PGCG. Although it is also important to maintain a high index of suspicion, discussion with family members should be tactful to prevent undue distress during the waiting period between differential diagnosis and definitive histopathologic diagnosis.

Because the clinical appearance of these various lesions can be remarkably similar, classification is based on their distinct histologic differences. The POF must be differentiated from the peripheral odontogenic fibroma (PODF) described by the World Health Organization.3

Histologically, the PODF has been defined as a fibroblastic neoplasm containing odontogenic epithelium.11

Despite a preponderance of literature supporting differentiation, some authors continue to argue that the POF (or peripheral cemento-ossifying fibroma) is the peripheral counterpart of the central cemento-ossifying fibroma.7

Endo et al attempted to distinguish cementifying fibromas from ossifying fibromas and fibrous dysplasias by using immunohistochemical analysis for keratin sulfate and chondroitin-4-sulfate in which the cementifying fibromas showed significant immunoreactivity for keratin sulfate and ossifying fibromas and fibrous dysplasias showed intensive immunostaining for chondroitin-4-sulfate.25

The term ‘cemento-ossifying’ is now regarded as outdated and scientifically inaccurate.26 because the clinical presentation and histopathology of cemento-ossifying fibroma are the same in areas where there is no cementum, such as the skull, femur and tibia. These are all ossifying fibromas; hence, those that occur in the jaws should not be termed cemento-ossifying fibromas only based of the presence of teeth. Moreover, there is no histologic or biochemical difference between cementum and bone. Cemento-ossifying fibroma is the term given mainly due to the presence of dysmorphic round basophilic bone particles within ossifying fibroma, which have arbitrarily been called cementicles. However, these so-called cementicles are not from cementum but instead represent a dysmorphic product of this tumor analogous to the keratin pearls, which are a dysmorphic product of squamous cell carcinoma.26

POFs contain areas of fibrous connective tissue, endothelial proliferation and mineralization. Endothelial proliferation can be profuse in the areas of ulceration, which can be misleading in clinical diagnosis, as the lesion may appear to be a pyogenic granuloma. The mineralized component of POF varies occurring in approxi-
Treatment consists of conservative surgical excision and scaling of adjacent teeth. The rate of recurrence has been reported at 8-20% in approximately 23, 35%, or 50 to 75% of cases according to published reports.

Mineralization can vary between cementum-like material, bone (woven and lamellar) and dystrophic calcification. Treatment consists of conservative surgical excision and scaling of adjacent teeth. The rate of recurrence has been reported at 8.9%, 9%, 16%, 14%, 16%, and 20% Therefore, regular follow-up is required. Local surgical excision including the involved periodontal ligament and periosteum of POF is the preferred treatment which was performed in this case.

The recovery was uneventful and there has been no recurrence seen during the 8 months follow-up period. Although POF is a benign, reactive lesion, the recurrence rate is fairly high. Therefore, the patient is still on regular follow-up.

CONCLUSION

POF is a slowly progressing lesion, with limited growth potential. Many cases will progress for long periods before patients seek treatment because of the lack of symptoms associated with the lesion.

Treatment consists of surgical excision, including the periosteum, and scaling of adjacent teeth. Close post-operative follow-up is required because of the growth potential of incompletely removed lesions and the possibility of recurrence (8-20%).

REFERENCES


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