CASE REPORT

Peripheral Odontogenic Fibroma (WHO-Type) – A Case Report

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Abstract

Introduction: Odontogenic fibroma is a rare benign tumor that is non-infiltrative, usually well-circumscribed and ectomesenchymal in origin. Like most odontogenic tumors, they are often presented as intraosseous/central variant contributing to 0.1% or an extra osseous/peripheral variant contributing to 1.2 – 4.7 % of all odontogenic tumors. They are composed of islands of odontogenic epithelium in the fibrous connective tissue stroma with varying amounts of dentin/cementum-like calcified material.

Case Presentation: The present case is of 41-year-old female patient who reported with an asymptomatic swelling in the right upper back tooth region since two months.

Management and Prognosis: Clinical diagnosis of pyogenic granuloma was made and the lesion was excised. Histopathological examination of the specimen showed ulcerated epithelium with highly cellular connective tissue stroma. Foci of irregular calcified material were seen along with odontogenic epithelial islands. Immuohistochemistry was performed for CK-14 protein which showed positive reaction.

Conclusion: The purpose of presenting this case is to highlight the importance of histopathology of every tissue submitted for arriving at a confirmatory diagnosis in addition to clinical findings.

Keywords: Fibroma, Gingival neoplasm, Odontogenic tumor, Reactive oral lesion

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Introduction

The odontogenic fibroma (WHO type) is a relatively rare, benign, gingival mass of fibrous connective tissue that can occur either as central or peripheral lesion, The extra osseous counterpart is designated as "Peripheral Odontogenic Fibroma (POF)".[1] In 2005 WHO classification, two histopathological subtypes of odontogenic fibroma were given - the epithelium rich (WHO-type or complex) and the epithelium poor type (simple type) that was later excluded in 2017 classification due to poorly defined and documented epithelial poor types.[2] Here, a well-documented case of Peripheral Odontogenic Fibroma (WHO- type) is presented.

Case Presentation

A 41-year-old female patient reported to the outpatient department with a chief complaint of swelling in the right upper tooth region for the past two months with no associated pain. Past medical and family history was non-contributory. Patient was apparently normal before 2 months after which she developed a swelling that is slow growing and attained a size of 1.5 x 1 cm after which she reported to the hospital. On extra oral examination, no relevant facial asymmetry was noticed. No lymph nodes were palpable.

On intraoral examination, a pedunculated mass on the right marginal and attached gingiva in relation to 13, 15, and 16 region measuring approximately 1.5 x 1 cm. Patient had undergone orthodontic treatment before 20 years for which she had extracted 14. The swelling was firm in consistency and non-tender on palpation. The swelling was pale pink in colour with reddish ulcerated areas (Figure 1). There was no evidence of pus discharge. Plaque and calculus deposits were present with generalized gingival inflammation. Intra oral periapical radiograph of the involved area revealed a horizontal bone loss (Figure 2). Based on these clinical and radiographic findings, provisional diagnosis of pyogenic granuloma was made. Differental diagnosis includes peripheral giant cell granuloma, pyogenic granuloma, hyperplastic gingival inflammation, peripheral odontogenic

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fibroma, traumatic fibroma and other gingival neoplasms. An excisional biopsy was performed under local anaesthesia as the immediate clinical concern was to excise and diagnose the gingival swelling. Secondly, flap surgery was planned for correction of the mucogingival defect in relation to molar and premolar.

Macroscopically, the gross specimen consisted of a soft tissue mass that was whitish-brown in colour, oval in shape, with an irregular border measuring about 1.2x2x1 cm, and firm in consistency (Figure 3). Histopathological examination revealed ulcerated surface epithelium with underlying connective tissue stroma. The underlying connective tissue stroma is highly cellular with a large number of plump, proliferating fibroblasts and bundles of collagen fibres. There were foci of irregular calcified material, as well as islands of odontogenic epithelial cells in the connective tissue stroma (Figure 4A&4B). Chronic inflammatory cell infiltrate consisting of lymphocytes, plasma cells, macrophages, extravasated RBCs, engorged blood vessels and budding capillaries were also seen. Based on these findings, diagnosis of Peripheral Odontogenic Fibroma (WHO type) was made. POF resembles both clinically and histologically with peripheral ossifying fibroma. The differentiating feature is that the former has a epithelial component derived from odontogenic origin where as the later is not. To confirm the odontogenic epithelial counterpart, immunohistochemistry was performed where the epithelial cells were immunoreactive to CK-14 protein (Figure 5).

The postoperative course was uneventful and follow up examination over one year have shown a satisfactory response with no significant sign of recurrence (Figure 6). Flap surgery was planned for correction of the mucogingival defect.

Discussion

The odontogenic fibroma (WHO type) is a relatively rare, benign, unencapsulated gingival exophytic mass of fibrous connective tissue. The World Health Organization (WHO) defined Odontogenic Fibroma as "a benign Odontogenic neoplasm of fibroblastic origin characterized by relatively mature collagenous fibrous tissue and varying amounts of odontogenic epithelium with potential to occur in either a central or an extra osseous location." POF is the name given to the extra osseous counterpart, being more prevalent by a ratio of 1.4:1.[1]

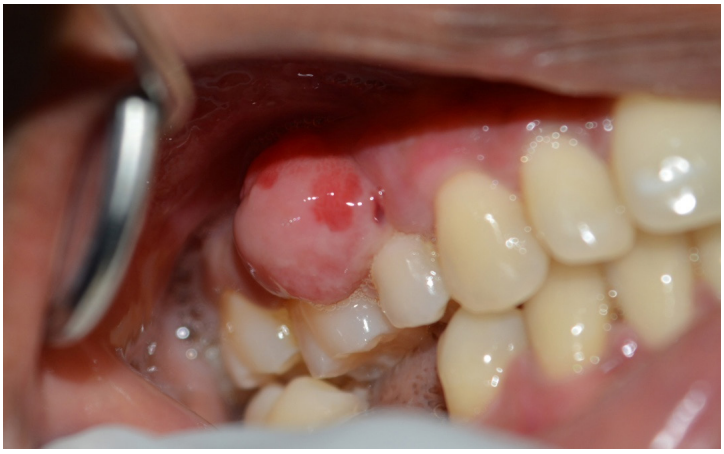

Fig. 1: Intraoral swelling in the right attached gingiva

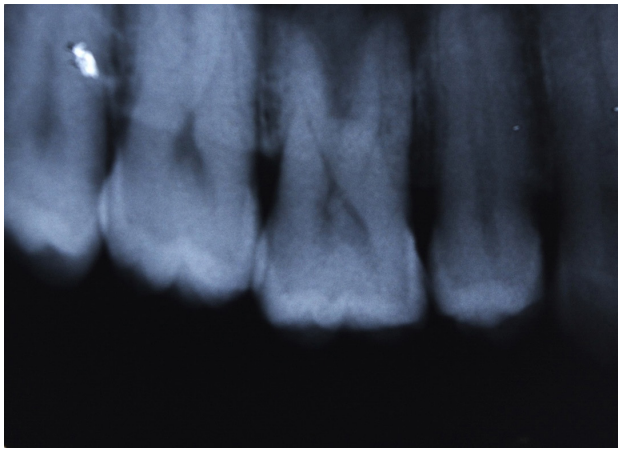

Fig. 2: Intraoral periapical radiograph of 15,16 and 17 showing horizontal bone loss

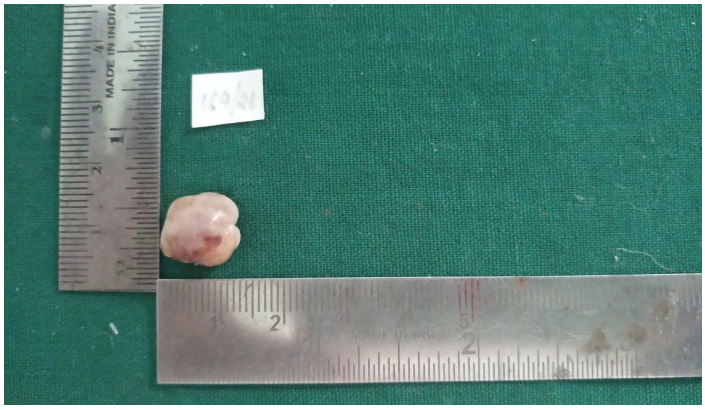

Fig. 3: Gross specimen

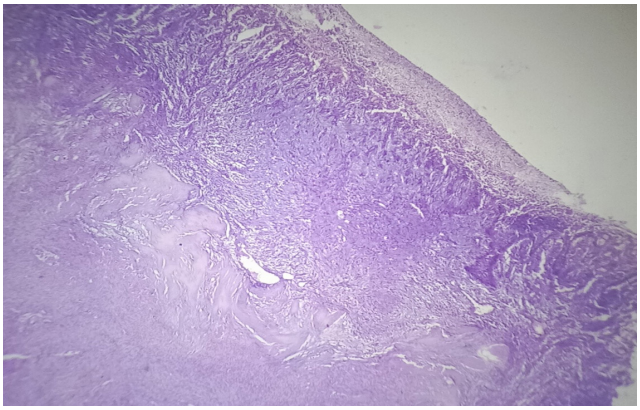

Fig. 4A: Ulcerated surface epithelium with underlying connective tissue stroma with plump, proliferating fibroblasts, collagen fibres and foci of irregular calcified material H&Ex100

POF was formerly confused with peripheral ossifying fibroma until it was defined as an Odontogenic tumor by the World Health Organization in 1992.[3]

The term POF was confused with that of peripheral ossifying fibroma until Gardner[4] made an attempt in clarifying the confusion between these terms. The prime distinguishing features between these two lesions are the differences in their connective tissue components.

POF's pathogenesis is still not fully understood. Based on the formation of these lesions in the dentulous areas, several investigators have suggested that the periodontal ligament is the source of the fibroblastic component. Wright conducted a histochemical analysis of eighteen cases of POF through in which he demonstrated the oxytalan fibres- a major part of the periodontal ligament, which supported the above statement.[5,6]

Ritwik and Brannon reported two cases in patients with edentulous mouth and stated that the periodontal ligament was not a viable source for the histogenesis of POF.[7] The epithelial component has its origins in the basal layer or dental lamina remnants. However, local factors such as inadequate restorations, presence of plaque or calculus, and excessive chewing forces might be possible etiologic factors for POF.[8]

Some studies also raised a hypothesis of considering peripheral odontogenic fibroma as a mixed tumor, since both epithelium and mesenchymal components are necessary for histological diagnosis.[9,10]

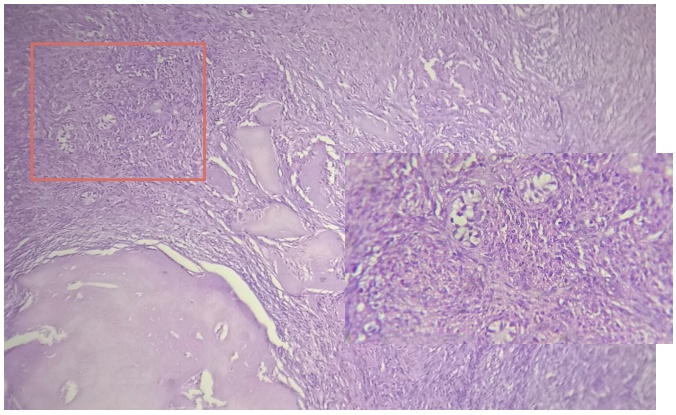

Fig. 4B: Foci of irregular calcified material resembling osteodentin. H&Ex100 Inset showing islands of Odontogenic epithelial cells. (H&Ex400)

The incidence of POF is said to be between 20 to 40 years with a slight predominance towards female gender as in our present case. It is a reactive lesion characterized by a growing nodule of soft tissue. It can be sessile or pedunculated and its appearance may vary from pink to red (ulcerated) and from smooth to irregular surfaces. Mineralisation's are commonly observed, resembling a cementum, bone/osteoid or dystrophic calcification.

Though it can arise in either arch, the tumor has been reported to have a slight mandibular predilection in the incisor cuspid region. In the present case the lesion was found in maxillary arch that was pedunculated, asymptomatic and firm in consistency. Radiographically, though POF does not involve the underlying bone and only rarely shows areas of calcification on radiograph, Ritwik and Brannon in their study, documented radiographic features in 12 cases out of the 151. The reported radiographic features include areas of calcification, superficial bone depression, and horizontal bone loss. Other features are alveolar bone resorption and tooth displacement. The present case shows no significant radiographic changes pertaining to the lesion.[7,9]

The histopathological findings are similar to central variant. Tumor consists of cellular fibrous connective tissue with the presence of islands/strands of Odontogenic epithelium that are scattered throughout the connective tissue. Dysplastic dentin, amorphous ovoid cementum like calcifications and trabeculae of osteoid may also be present.[11]

CK14 is the main intermediate filament of odontogenic epithelium, which is observed in the dental lamina, in the reduced enamel epithelium and in almost all cells of the enamel organ. Its absence in regions of advanced amelogenesis may suggest loss of CK14 as a consequence of the cellular secretory activity.[12] Antibodies against the cytokeratins have been utilised to define the tumour and understand histogenesis. Cytokeratins have shown beneficial in identifying odontogenic epithelium in numerous biomedical researches, and as a result, they have helped in the identification of various neoplasms and cysts when an odontogenic origin is suspected.[13] Odontogenic tumors with epithelial component frequently express CK 14 and 19. Numerous studies have been done to report the expression of Cytokeratin 14 in various odontogenic tumors.

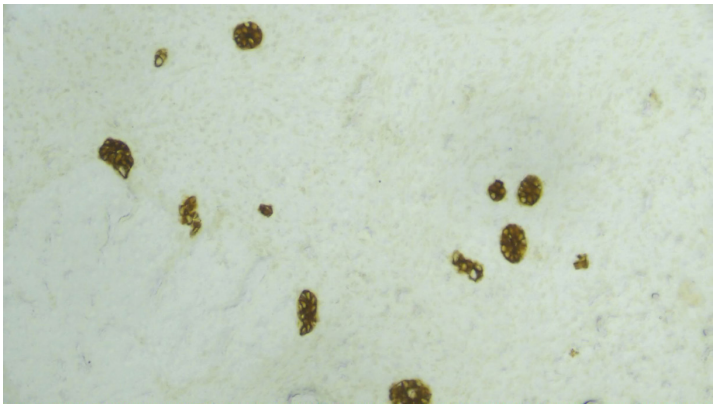

Fig. 5: Immunohistochemical staining showing positivity for CK-14 protein. IHC x400)

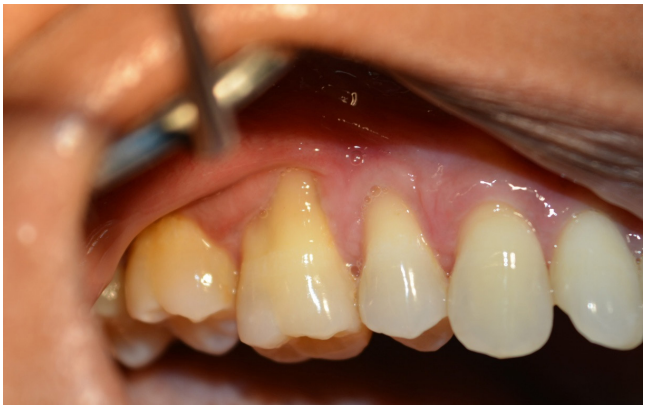

Fig. 6: Postoperative image showing no recurrence after one year followup

Thus, CK 14 can be used as a marker for identifying the odontogenic epithelial origin of tumor.[14] Speight et al in his review mentioned about IHC in 14 cases in which all the cases exibited the positive expression to cytokeratins like AE1/AE3, CK 14 and CK 19. The epithelial islands were immunoreactive to CK 14 protein in this case which further aided and supported the epithelial counterpart of the tumor.[15]

The POF is usually treated by local surgical excision and the prognosis is good. The frequency of recurrence rate of POF varies greatly and has been reported to range from very low to as high as 38.9% - 50%.[8] However in his study Ritwik et al stated that the presence of calcification in apposition to odontogenic epithelial remains and basal cell layer budding of the surface stratified squamous epithelium were histologic factors that were statistically significant for conjunction with POF recurrence. Apposition of calcification and odontogenic epithelial rests was related with a lower recurrence, while basal cell layer budding was associated with a higher recurrence.[7] Complete surgical removal of the tumor aids in preventing the recurrence.

Conclusion

To conclude, although this lesion is rare and benign in nature, pathologists and clinicians should not omit this lesion when observing a growth in the oral cavity. Any asymptomatic nodule in the gingiva must include POF as a part of diagnosis. The definitive preoperative diagnosis should be carried out with a biopsy and histopathologic investigation in order to choose the appropriate treatment option for the patient and a long-term follow-up is required after surgery to rule out any recurrence.

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