

## Peripheral odontogenic fibroma an uncommonly overlooked benign neoplasm: A case report and review of literature

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### ABSTRACT

Peripheral odontogenic tumors (POTs) are one of the rare neoplasms to occur on the gingiva. Peripheral odontogenic fibroma (POdF) is the most common POT constituting a prevalence of 1.2%. A plethora of lesions sharing similar clinical features makes the diagnosis difficult. Histopathological examination plays a key role in these types of cases. Here, we present a rare case of POdF in a 36-year-old female patient with a lobulated pebbled-like exophytic growth on the right gingiva. Histopathological investigation showed the presence of odontogenic epithelium and dystrophic calcification which are the pathognomonic features of POdF. After surgical removal, POdF is seen to recur, but the exact recurrence rate cannot be estimated due to the scarcity of reported cases. In this case, follow-up of the patient showed no recurrence at 6, 12, and 18 months post-surgery.

**Key words:** Gingival carcinoma, Neoplasm, Peripheral odontogenic fibroma, Peripheral odontogenic tumors

Peripheral odontogenic tumors (POTs) are benign tumors seen as an outgrowth of oral soft tissue, commonly on the gingiva. They have histological features similar to their central/intraosseous counterparts but are found in the mucosa overlying the alveolar region of the jaw. Peripheral odontogenic fibroma (POdF) is the most common POT [1]. According to the World Health Organization (WHO), they show a prevalence rate of 1.2% among all odontogenic cysts and tumors [2]. Among POT, POdF is more frequent than its central analogue [2]. The WHO has defined POdF as “a fibroblastic neoplasm containing varying amounts of apparently inactive odontogenic epithelium. It may contain dentine and/or material resembling cementum.” This peripheral variety is uncommon and represents clinically as a focal swelling.


### CASE REPORT

A 36-year-old female patient visited the Department of Oral Medicine and Radiology complaining of outgrowth in her lower right back region of the jaw for 14–15 months. The outgrowth was gradual in onset without any other symptoms until the past 2 months, when she started experiencing pain in a similar region. The pain was dull aching in nature, intermittent and used to aggravate upon mastication. She had no relevant family and

medical history. She visited a dentist 1 year ago regarding the same, where she was advised to get the tooth removed in the concerned area as it was thought that the outgrowth would regress after the extraction. However, after the extraction, outgrowth rapidly increased in size.

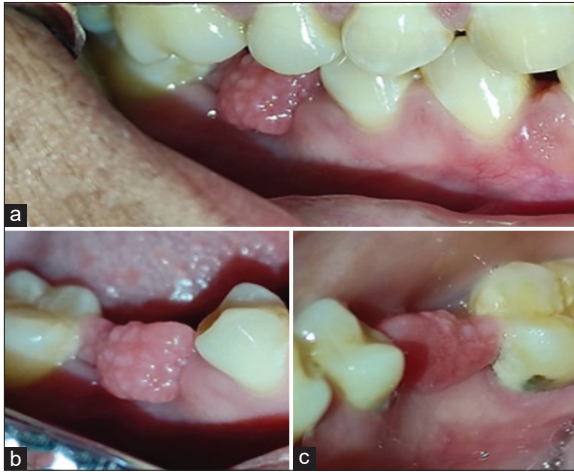
On examination, all vitals were in the normal range. The history of tobacco consumption in any form was negative. The patient mentioned that she applied digital pressure over the outgrowth as she thought that it would regress the lesion but she did not notice any change. On intraoral examination, a small 1 cm × 0.5 cm solitary, well-defined, and dome-shaped overgrowth was present on the right alveolar ridge in the edentulous region of 46 (Fig. 1a). The overlying mucosa was intact with a pebble-like appearance on the buccal surface (Fig. 1b), while the lingual surface was smooth and slightly erythematous (Fig. 1c). On palpation, it was firm in consistency and lobulated on the buccal surface and smooth on the lingual surface with slight tenderness. The base was slightly pedunculated and was mobile to a certain extent.

Extraoral palpation revealed right submandibular lymphadenopathy. Depending on the clinical findings, a provisional diagnosis of gingival squamous cell carcinoma was suspected and a periapical radiograph was advised which showed a healing socket with no other significant findings. Hematological investigations were done which were found to be within normal limits. An excisional biopsy was performed under local anesthesia (Fig. 2).

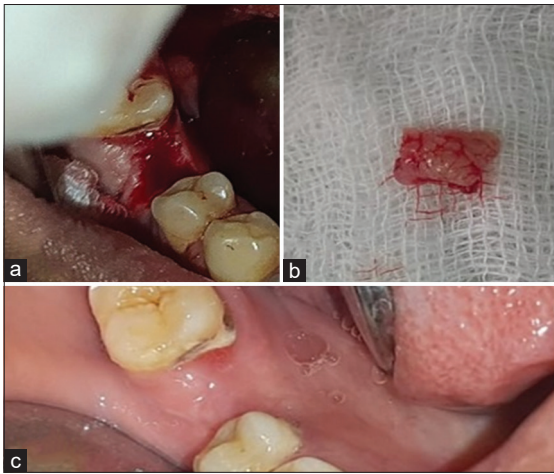
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**Figure 1:** (a) Outgrowth in mandibular right posterior region; (b) buccal view showing pebble-like appearance; and (c) lingual view showing smooth and erythematous

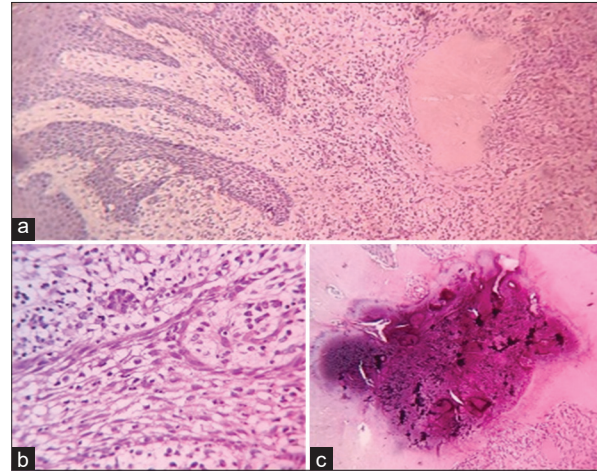


**Figure 2:** (a) Immediate post-operative view; (b) excised gingival tissue; and (c) 15 days post-operative view

Histopathological examination revealed parakeratinized stratified squamous epithelium and slender rete ridges with the plump fibroblast, collagen fiber bundles, and infiltration of inflammatory cells. The islands of odontogenic epithelium along with ossification were seen in many places. Dystrophic calcification was evident in one section (Fig. 3). On the basis of this, a diagnosis of POdF was confirmed. Follow-ups of the patient showed no recurrence at 6, 12, and 18 months post-surgery.

## DISCUSSION

POdF is one of the rare benign neoplasms of ectomesenchymal origin [3]. It is classified into two types: The WHO type contains inactive odontogenic epithelium [2], whereas, the non-WHO type is devoid of epithelium [4]. It is a rare neoplasm, being considered as the extrasosseous analog of the central odontogenic fibroma characterized by different degrees of odontogenic epithelium embedded in a mature, fibrous matrix [3]. In the past, it was designated as odontogenic gingival epithelial hamartoma due to epithelial component rather than the fibroblastic component of POdF. However, since they are developmental in nature and



**Figure 3:** H&E stain shows (a) slender and elongated rete-pegs; (b) fibroblast proliferation with odontogenic epithelial islands; and (c) dystrophic calcification

develop after completion of the dentition, this term is no longer used [5].

The incidence rate of the lesion is seen to be highest in the second and fourth decades and it occurs twice as often in females [4-6]. A case series of 151 patients carried out by Ritwik and Brannon found patient's ages ranging from 5 to 83 years with a mean age of 37.3 years [6]. The literature indicates that the occurrence of POdF is very rare in children [5].

The most common location was the attached gingiva, usually in the molar/premolar area with even distribution in both jaws [7]. However, a study done by Eversole showed that it arises frequently in the anterior gingival regions [4].

It usually occurs in form of exophytic sessile/pedunculated mass arising from the interdental region [4,6,7], which may present on the gingiva and alveolar ridge. It may appear as red, inflamed, and ulcerated or coral pink, fibrous, and firm mass. A study carried out by Ritwik and Brannon reported the duration of the lesion varies from 3 weeks to 20 years [6]. Radiographic features comprising calcifications and crestal alveolar bone resorption is rare, but in some instances, they might be seen [4,6].

Microscopically, these lesions are composed of fibrous stroma showing a varying degree of collagen arranged from loose, myxoid patterns to mature collagen fibers with low-to-moderate cellularity. Odontogenic epithelial elements arranged in clusters, nests, and cord-like fashion and bone, cementum-like, or dystrophic calcifications may be dispersed throughout the fibrous component [1,5].

Various benign and malignant gingival neoplasm and inflammatory conditions show overlapping clinical features and can be considered in the differential diagnosis. Gingival squamous cell carcinoma appears as an exophytic growth with a granular to the papillary or verrucous surface or an ulcerative lesion. It is rarely associated with tobacco consumption and gingival pain is the classic symptom. Similarity to common periodontal lesions might leads to misdiagnosis [8] Inflammatory lesions such as fibrous hyperplasia, fibroma, peripheral granuloma, peripheral giant cell granuloma (PGCG), and peripheral ossifying fibroma

(POF) are considered in the differential diagnosis of POdF [9]. Clinically, POF is a well-defined, pink to red mass, present on the gingiva, with either a sessile or pedunculated base. The surface may be intact or ulcerated. Histologically, POFs show prominent areas of intensely cellular connective tissue with foci of bone, osteoid, or other calcifications [9]. POF is distinct from the POdF in histologic appearance as it lacks epithelium. Pindborg *et al.* defined it as “a fibroblastic neoplasm containing varying amounts of odontogenic epithelium” [10]. The PGCG derives from the periodontal tissues; clinically, it appears as a reddish-purple nodule. It has the highest incidence in the 5<sup>th</sup>–6<sup>th</sup> decades, whereas, POdF is mostly seen in the 2<sup>nd</sup>–4<sup>th</sup> decades and is commonly presented in the posterior area. The PGCG shows characteristic hemorrhage which leads to hemosiderin deposits. Numerous multinucleate giant cells and spindle cells are present. Although POdF has multinucleated giant cells, they are lesser in number compare to PGCG also odontogenic epithelium with abundant fibroblast in its collagenous stroma that is a diagnostic feature of POdF [5]. Oral pyogenic granuloma is associated with a wide age range and slight female predominance. Gingiva is the predominant site but can be seen in other parts. Clinically, it occurs as a sessile to elevated mass which is deep red to reddish-purple in color, soft, and painless. Microscopically, it is covered by non-keratinized or parakeratinized stratified squamous epithelium. The bulk of the lesion contains the mass of angiomatous tissue mostly solid endothelial and capillary-sized blood vessels proliferation. Collagen is usually sparse. If the surface is ulcerated, edema is prominent containing plasma cells, lymphocytes, and neutrophils [11].

POdFs are treated by complete surgical excision followed by a biopsy. Some literature mentioned no recurrence [4]. However, if present, it is based on histologic variants. Higher recurrence is seen when there is the presence of the surface epithelium with basal cell layer budding, whereas, a lower rate of recurrence is seen in case of calcifications in juxtaposition to odontogenic epithelial rests are present [6].

## CONCLUSION

Odontogenic fibroma is one of the benign odontogenic tumors occurring centrally and peripherally in the bones and on the gingiva, respectively, the latter being relatively more common. As POdF is a rare entity with diverse clinical presentation resembling

other gingival lesions, it can be misdiagnosed clinically as inflammatory or even a malignant lesion. An accurate diagnosis with the help of pathognomonic features is of utmost importance and histopathological investigation plays a key role to rule out the nature of the lesion as it has significant growth potential also chances of recurrence have been seen associated with it. In this case, a unique pebbled-like appearance associated with areas of dystrophic calcification and pain were characteristic features.

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