Recurrent Peripheral Odontogenic Fibroma: A Case Report with the Review of Literature
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ABSTRACT
Peripheral odontogenic fibroma (POF) is a rare benign mesenchymal odontogenic neoplasm with or without odontogenic epithelium, which is considered to be the mucosal analogue of central odontogenic fibroma. Though peripheral odontogenic tumours are rare, comprising of only 0.05% of all biopsy specimens, POF is the most common peripheral odontogenic tumour, more common than its central counterpart by a ratio of 1.4:1. In this report, we describe a healthy 40-year-old female who was seen with a recurrent POF of the left mandible.

KEYWORDS: Recurrent peripheral odontogenic fibroma, Extrasosseous variant, Biopsy specimens, World health organization, Benign neoplasm, COF

INTRODUCTION
The odontogenic fibroma (OF) is a rare, representing 1% to 2.5% of oral biopsy specimens. It occurs either as a central (intraosseous) lesion (COF) or in a peripheral location (POF)¹. The World Health Organization (WHO) defined it as ‘a benign odontogenic neoplasm of fibroblastic origin characterised by relatively mature collagenous fibrous tissue and varying amounts of odontogenic epithelium with potential to occur in either a central or an extrasosseous location. The extrasosseous counterpart being designated as peripheral odontogenic fibroma’². The POF presents as an elevated exophytic lesion with no radiological changes in the underlying alveolar bone. The age of diagnosis varies, with peak incidence in the third and fourth decades of life. The most common site for POF is the canine/premolar area both in the mandible and maxilla³⁴.

The odontogenic fibroma comprised 2.9% of the total of 1133 lesions and occurred more frequently as a peripheral than as an intraosseous lesion¹. Daley and Wysocki have reported POF to be the third most common odontogenic tumour and the most common peripheral odontogenic tumour⁵. Though its biological behaviour is uncertain, it is considered as a benign neoplasm with an appreciable recurrence rate⁶.

CASE REPORT
A 40 year old female patient reported to I.T.S Dental College Greater Noida with a chief complaint of swelling in lower front tooth region since one and half year. Patient was apparently asymptomatic 3 years back when she first noticed swelling which was painless and growing in size. The growth was excised at a private dental college but it reoccurred at the same site one and half year back and had reached to the present size. Patient had no relevant medical history. On examination, a single sessile swelling 2.5 x 1.5 cm in size, firm in consistency, reddish in colour was present in left lower

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front tooth region, extending from mesial surface of 31 to distal surface of 34, superiorly the swelling was extending up to the occlusal level of 32 and 33 and inferiorly the lesion extending to the vestibule (Figure 2). Radiographic examination did not demonstrate any bony destruction (Figure 3). An excisional biopsy was performed under local anaesthesia. Gross examination reveals multiple pieces of soft tissue and teeth with attached soft tissue. Soft tissue was firm in consistency whitish black in colour measuring 3 x 1 cm, 0.4 x 0.3 cm, 0.2 x 0.3 cm, 0.2 x 0.4 cm, 0.2 x 0.2 cm (Figure 4). Microscopic evaluation revealed parakeratinised stratified squamous epithelium with slender, deep penetrating rete ridges (Figure 5) with areas of epithelium exhibiting budding or off shots (Figure 6). Underlying connective tissue stroma was highly cellular with plump fibroblast and with areas of inactive odontogenic epithelium arranged in the form of nests and strands along with scattered amorphous calcifications (Figure 7). Chronic inflammatory cells and capillaries were also present. A final diagnosis of peripheral odontogenic fibroma was made.

DISCUSSION

The POF has been widely accepted as an odontogenic tumour of mesenchymal origin. The epithelial component of it has been considered inactive despite the fact that in some lesions the epithelium is abundant and occasionally the dominating feature that has led some investigators to question the possibility of a mixed odontogenic origin for the lesion[7].

The exact aetiology of POF is unknown. Many people have suggested that it is a neoplastic process; while others have argued that it is reactive, arising from the periodontal ligament or dental lamina. Local chronic irritants such as poor restorations, ill-fitting dentures, calculus and plaque, heavy masticatory forces and external trauma are possible etiologic factors for the POF[8].

It appears as a firm, slow-growing and usually sessile gingival mass covered by normal appearing mucosa[9,2]. Clinically, it cannot be distinguished from common fibrous gingival lesions. The lesion is generally elevated and non-ulcerated clinically and non-encapsulated microscopically[2]. Our case presented clinically as a single, non-ulcerated swelling which was covered by normal mucosa, approximately 2.5 x 1.5 cm in size and firm in consistency.

The age range of patients with peripheral odontogenic fibroma varies from 5 to 83 years with a mean age of 37.3 (17.8) years. The highest incidence of the lesion was in the second and fourth decades. There is equal gender distribution, with a slight female predilection[9]. It can arise throughout either arch but tends to occur in the mandibular canine-premolar and maxillary anterior areas[7]. The age of the patient in present case also coincides with that reported in the literature, i.e., 40 years. Our case also occurred in the most common site i.e. the mandibular canine region.

Radiographically, peripheral odontogenic fibroma does not involve the underlying bone and only sometimes shows areas of calcification on radiograph[2]. The same is applicable in our case where there was no bony involvement.

The histologic spectrum of this lesion is wide. The connective tissue ranges from loose (almost myxomatous) to markedly cellular or to relatively a cellular and well-organised stroma. Islands or strands of odontogenic epithelium are seen scattered throughout connective tissue, which may be either prominent or scanty. Dysplastic dentin, amorphous ovoid cementum like calcifications and trabeculae of osteoid may be present. Peripheral odontogenic fibroma shows histopathologic features similar to those of the central odontogenic fibroma (WHO) type[2]. Histopathological evaluation of our case confirmed the findings of peripheral odontogenic fibroma.

Differential diagnosis includes peripheral ossifying fibroma, peripheral ameloblastoma inflammatory fibrous hyperplasia, epulis, pyogenic granuloma, peripheral giant cell granuloma or gingivitis[7].

Peripheral ossifying fibroma is a reactive fibrous lesion of the gingiva, unlike the POF. As a result of marked
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Figure 1: Extra-oral view showing no facial symmetry

Figure 2: Intra-oral view showing a single swelling, reddish in color, firm in consistency

Figure 3: OPG did not demonstrate any bony destruction

Figure 4: Gross examination of specimen

Figure 5: Photomicrograph showing surface squamous epithelium exhibiting slender, deep penetrating rete ridges

Figure 6: Photomicrograph showing strands of odontogenic epithelium budding from the overlying epithelium

Figure 7: Photomicrograph showing islands of odontogenic epithelium in a moderately cellular fibrous stroma

Figure 8: Photomicrograph showing extracted teeth associated with lesion
epithelial proliferation, the POF may be confused histologically with peripheral types of ameloblastoma or of calcifying epithelial odontogenic tumour. The epithelial islands of the POF are smaller than those of the ameloblastoma and do not exhibit such features of the basal cell layer, as hyperchromatism, intracytoplasmic vacuoles, and polarisation of the nucleus away from the basement membrane, as found in various degrees in that tumour. Additionally, hard tissues, such as dentin or cementum-like are not found in ameloblastoma[10].

The recommended treatment for the POF is removal of irritating factors and simple surgical excision, which should include the periodontium and the periosteum, to reduce the chance of recurrence. The recurrence rate after simple surgical resection has been reported to be between 8% and 20%[8].

Kenney et al. reported 1 of 13 cases recurred, and deVilliers Slabbert and Altini found 1 of 30 recurred. In 1987, Buchner et al. had no recurrences in 4 cases with follow-up. On the other hand, Daley and Wysocki found 7 (38.9%) of 18 cases recurred and Garcia et al. found 3 (17.6%) of 17 cases recurred[5].

The recurrence of POF is associated with 2 histologic variables:

- Presence of basal cell layer budding of the surface epithelium is associated with a significantly higher recurrence rate of this lesion.
- Presence of calcifications in apposition to odontogenic epithelial rests is associated with significantly lower recurrence rate[9].

This case demonstrated the importance of radiographic examination of all exophytic gingival lesions to exclude a central counterpart and of histological examination of the biopsy specimens to establish the exact diagnosis.

**CONCLUSION**

This case demonstrated the potential for recurrence of peripheral odontogenic fibroma. Periodic evaluation of patient by clinical and radiographic examination is essential.

**REFERENCES**