CASE REPORT

Peripheral cemento-ossifying fibroma – A case report

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Abstract
Peripheral cemento-ossifying fibroma (PCOF) is an osteogenic neoplasm which is rare and is usually present as an epulis-like growth. It is of a reactive nature rather than a neoplastic growth. Its pathogenesis is uncertain. Predominantly, it affects adolescent and young adults with greatest prevalence around 20–30 years. Here, we report a rare clinical case of PCOF of the mandible with a history of just 2 months, occurring in a 23-year-old male involving the dentulous mandibular anterior region.

Keywords: Cementifying fibroma, gingival overgrowth, granuloma, irritational fibroma, peripheral cemento-ossifying fibroma

Introduction
The peripheral cemento-ossifying fibroma (PCOF) is a neoplasm which is osteogenic in nature.\(^1\) It is believed that PCOF arises from the periodontal ligament. It accounts for 9.6% of all gingival lesions and 3.1% of all oral tumors. Maxillary anterior region is the most frequently involved site. It mostly occurs in the teenagers and young adults and has a female predilection. Trauma, ill-fitting dentures and faulty restorations, local irritation due to plaque and calculus, and hormonal disturbances are the known predisposing factors. An early diagnosis and surgical excision is required to prevent its recurrence. There are many types of localized reactive lesions that may occur on the gingiva, including pyogenic granuloma, peripheral giant-cell granuloma, focal fibrous hyperplasia, and PCOF. These lesions may arise as a result of irritants such as microorganisms, trauma, plaque, calculus, dental appliances, and dental restorations.

The World Health Organization classification in 1992 includes two histologic types, namely, cementifying fibroma and ossifying fibroma that may be clinically and radiographically indistinguishable under a single designation of cemento-ossifying fibroma.\(^2\)

PCOF is commonly seen in young adults and female adolescents with the peak age ranging between 10 and 19 years as a gingival growth near the interdental papilla of the maxillary incisors or canines. It usually appears as a nodular mass, either sessile or pedunculated. The color of the growth ranges from red to pink, and the surface is usually smooth and seldom ulcerated.\(^3\) The lesion is usually present for many months to years before it is diagnosed. The diagnosis is difficult on the basis of clinical features; hence, a histopathological examination is mandatory for its confirmation.\(^4,5\)

Case Report
A 23-year-old male patient presented at the Outpatient Department of Oral Medicine and Radiology with the chief complaint of gum swelling in the lower left front tooth region [Figure 1]. The swelling had been present for 2 months and had been slowly increasing in volume over that time. Occasionally bleeding occurred when the patient brushed his teeth and was associated with slight pain. He denied tobacco and alcohol use. The patient had given a history of handicapped since birth (defect in both lower limbs) and the patient’s mother revealed the history of the patient’s slow motor function (delayed developmental milestones) and low intelligence quotient.

Family history revealed that his parents had consanguineous marriage. The patient’s dental history was unremarkable.
Clinical examination

Extraoral examination showed facial symmetry and the overlying skin showed no signs of inflammation. The regional lymph nodes were neither enlarged nor tender. Intraoral examination revealed a solitary sessile growth, approximately 1.5 cm × 1 cm in diameter, roughly ovoid in shape, and color of the mucosa overlying the growth was normal, non-tender. The growth was confined to the labial gingiva in the mandibular left lateral incisor and canine region [Figure 2]. The lesion was neither fluctuant nor did it blanch with digital pressure, and it was firm in consistency. The overlying mucosa was smooth and there was no visible pulsation or discharge. The lingual gingiva was not involved. The local irritants, i.e., plaque and calculus, were abundant in the 32, 33, and 34 region.

A provisional diagnosis of irritational fibroma was made, and pyogenic granuloma, peripheral odontogenic fibroma (PODF), and peripheral giant-cell granuloma were considered as the differential diagnoses.

Radiographic examination

Intraoral periapical radiograph was obtained [Figure 3]. The radiographic examination showed horizontal bone loss till middle 3rd of the root in 31, 32, and 33 with widening of PDL space in 32.

Blood investigation

The patient underwent blood investigation before the excisional biopsy and all readings including hemoglobin, bleeding time, and clotting time were within normal limits.

Treatment

The lesion was completely excised [Figure 4] under local anesthesia. Subsequently, a histopathological examination of the specimen was performed.

Microscopic examination

The H and E stained soft tissue section showed parakeratotic stratified squamous epithelium overlying fibrocellular connective tissue. The connective tissue is predominantly fibrous with spindle-shaped cells. Focal areas of irregular immature bone trabeculae with osteoblastic rimming and osteocytes in lacunae are present. Numerous cementoid areas are seen. Mild inflammatory infiltrate composed of lymphocytes and plasma cells are also present.

The final diagnosis was given as PCOF.

Follow-up

The patient presented for follow-up examination 7 days postoperatively. The surgical site appeared to be healing well.

Discussion

Montgomery coined the term “PCOF” in 1927. PCOFs had been described in the literature. There are many synonyms that have been given to similar lesion such as peripheral fibroma with calcifications, epulis, peripheral cementifying fibroma, peripheral ossifying fibroma, peripheral fibroma with cementogenesis, calcifying fibroblastic granuloma, and PCOF. According to the WHO 1992 classification, PCOF is considered as an osteogenic neoplasm, which is frequently associated with irritant agents such as bacterial plaque,
calcium, orthodontic appliances, irregular restorations, ill-adapted crowns, and hormonal disturbances. Although the origin of the lesion is uncertain, it is said that it originates from cells of the periodontal ligament because to the following reasons:

a. Involvement of the gingiva exclusively (mainly interdental papilla)
b. The vicinity of the gingiva to the periodontal ligament and
c. Some lesions showed the presence of oxytalan fibers within their mineralized matrix.

In the present case report, it is assumed that local irritating factors such as plaque and calculus played a causative factor. PCOFs show increasing occurrences in the second decade and declining incidence after the third decade. 

Very few cases in the elderly have been reported to date, for example, studies by Bhasin et al., Dalghous and Alkhabuli and Mohiuddin et al. The site of occurrence is the interdental papilla of the maxillary incisor - canine region, whereas, in the present case, the lesion was observed in the left mandibular lateral incisor - canine region, which is not a common site. However, similar cases with lesions in the mandibular region have been reported by Yadav and Mishra and Passos et al. and those in edentulous regions have been reported by Kumar et al. and Yokoyama et al. The clinical presentation of PCOF is as a sessile or pedunculated exophytic mass, measuring about <2 cm in diameter (occasionally >10 cm). The color of the growth is mostly similar to that of the mucosa unless the surface is ulcerated. Sometimes, it may be associated with migration of the adjacent teeth. It is mostly a slow-growing mass that takes many months to years to reach a diagnosable stage, as seen in this case, the history of a short duration of 2 months was given by our patient and his family, which was quite uncommon. There are cases with such a short histories of about 3 months of duration of the tumor, which have been reported by Trasad et al., Satish et al., and Sudhakar et al. Radiographically, PCOF exhibits a well-defined, mixed, and radiolucent-radiopaque lesion, along with calcified material. Others may show a wispy or flocculent pattern. Radiographs of the present case revealed a horizontal bone loss with widening of periodontal ligament with respect to the left mandibular central incisor, lateral incisor, and canine. Clinically, when a case presents with a gingival lesion, it is very crucial to establish its differential diagnosis, which in the present case can be irritational fibroma, peripheral giant-cell granuloma, or pyogenic granuloma. As these lesions can be remarkably similar, therefore, classification is based on their distinct histological differences. It is crucial to differentiate PCOF from the PODF and must be classified as described by the WHO. PODF has been histologically defined as a fibroblastic neoplasm with an odontogenic epithelium. Although there is a significant number of literatures which support the differentiation, some authors continue to consider that the PCOF is the peripheral counterpart of the central cemento-ossifying fibroma.

The management includes an elimination of the associated local irritating factors, oral prophylaxis, and total surgical excision, accompanied by the involved periodontal ligament, and periosteum, to reduce the possibility of recurrence, as the recurrence rate is as high as that of approximately 8–20%. Therefore, a long-term follow-up is also essential postoperatively. The patient reported here is on periodic postoperative follow-up.

Conclusion

The cemento-ossifying fibroma is a relatively rare lesion and is considered as an osteogenic tumor (non-odontogenic) with variable clinical manifestations. It is defined as a well demarcated mass, with a fibrous tissue and containing variable amounts of mineralized material resembling bone (ossifying fibroma), cementum (cementifying fibroma), or both. It is mostly a slow-growing mass, which takes months to years to reach to a diagnosable stage, as opposed to the history of a short duration of merely 2 months given by our patient, which is quite unexpected. The common site of occurrence is the interdental papilla of the maxillary incisor - canine region, whereas, in the present case, the lesion has been observed in the left mandibular incisor - canine gingival region, which is a less common site. Radiographically, horizontal bone loss with widening of periodontal ligament space was evident. Thus, in the present case report, we have described a unique case of PCOF in a 23-year-old male, with uncommon features.

References
