

A Rare Case Report of Coexistence of Peripheral Ossifying Fibroma with Fibrolipoma

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ABSTRACT

Introduction: Peripheral ossifying fibroma belongs to the spectrum of reactive gingival hyperplasias. Although it is believed to occur as a gingival overgrowth in response to local irritants, the pathogenesis of this lesion is still a controversy.

Aim: This case report highlights the presence of a rare combination of coexistence of peripheral ossifying fibroma (POF) and fibrolipoma.

Case report: A case of POF in a 65-year-old female has been presented with its detailed clinical, radiographic, and histopathological findings along with the diagnostic challenges associated with such lesions and the management approaches for the same.

Conclusion: Establishing an early diagnosis becomes imperative to rule out the potential for malignant transformation. This case presents a rare combination of coexistence of fibrolipoma in buccal mucosa which in itself is a rare finding.

Keywords: Benign tumor, Fibrolipoma, Lipoma, Peripheral ossifying fibroma, Reactive lesion.

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INTRODUCTION

Gingival tissues are highly susceptible to developmental changes in the form of various reactive and neoplastic lesions. Peripheral ossifying fibroma is an inflammatory growth of the gingiva developing in response to plaque and other associated local factors favoring plaque accumulation and originating from the cells of the gingival corium, periosteum, and periodontal ligament (PDL). Other terms used in reference to POF are peripheral cementifying fibroma, peripheral fibroma with cemen-

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togenesis and peripheral fibroma with osteogenesis, peripheral fibroma with calcification, calcified or ossified fibrous epulis, and calcified fibroblastic granuloma. The sheer number of names used for fibroblastic lesions indicates that there is much controversy surrounding the classification of these lesions. However, when bone predominates, "ossifying" is the appellation, while the term "cementifying" has been assigned when trabeculae or spheroidal calcifications are encountered. When the bone and cementum-like tissues are observed, the lesions have been referred to as cemento-ossifying fibroma.²

On the contrary, fibrolipoma of the oral cavity is a rare entity among them, with only 35 cases reported in the literature. Extraorally, fibrolipomas have been reported in the esophagus, pharynx, colon, trachea, and larynx. Intraorally, they can occur at various sites, such as buccal mucosa, lips, tongue, palate, buccal vestibule, floor of the mouth, and retromolar area. A case of an POF coexisting with intraoral fibrolipoma in the buccal mucosa, a rare combination, is discussed here.

CASE REPORT

A 65-year-old female patient came with the chief complaint of growth in lower right front region of jaw since 4 to 5 years and food lodgment in the same region since 1 year.

Patient was apparently alright 4 to 5 years back, when she noticed a pea-sized growth in the right front region of lower jaw, which very slowly grew to the present size. As the growth was asymptomatic, she never consulted any physician. As the growth increased in size, food lodgment started occurring since a year and she came for dental consultation. She gave a history of cataract operation 4 years back and piles operation 1 year back for which she was admitted for 1 week. She had a habit of mishri application twice a day and spat it out after 30 minutes. Patient does not follow any other oral hygiene maintenance regimen. She was moderately built, of normal gait and posture. Vital signs were normal.

Extraoral examination showed lip incompetence due to an intraoral gingival growth in the lower anterior region of jaw. Intraoral examination showed the presence of $30 \times 20 \times 15$ mm nodular pedunculated growth i.r.t attached gingiva and involving the interdental papilla of 41 to 45. The overlying mucosa looked pale, smooth with no presence of a sinus tract (Fig. 1). On palpation, it was nontender,

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Fig. 1: A 30 × 20 × 15 mm pedunculated growth wrt attached gingiva of 41 to 45



Fig. 2: A 10 \times 7 mm soft tissue fibrotic growth on right buccal mucosa just below the line of occlusion, 10 mm posterior to right buccal commissure



Fig. 3: An occlusal radiograph showing calcification approximately 3 mm × 4 mm projecting perpendicularly between 43 and 44

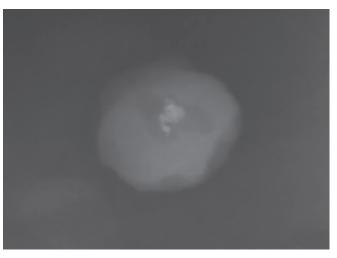


Fig. 4: Radiograph of the excised section showing calcification in the center surrounded by a radiolucent halo

firm to hard in consistency, and noncompressible nodular growth. The lower anterior teeth were vital and nonmobile with severe crowding. On intraoral examination of the soft tissues, there was another small nodular soft tissue mass in the right buccal mucosa 1 cm posterior to angle of mouth at the occlusal level in relation to 45, 44 area. The pedunculated mass was oval in shape, well-circumscribed and about 10×7 mm in dimension. It extended anteroposterior from distal aspect of 44 to the mesial of 45 in the buccal mucosa (Fig. 2). The mass was pale pinkish yellow in color with a smooth surface with no sign of any discharge or bleeding. It was nontender, soft, and compressible on palpation. The adjacent teeth were nontender to percussion with sharp cusps due to severe attrition. Periodontal status was very poor.

So, based on history and the clinical presentation, provisional diagnosis of POF and traumatic fibroma w.r.t attached gingiva of 41 to 45 and w.r.t right buccal mucosa was considered respectively. Peripheral giant cell granu-

loma (PGCG), peripheral fibroma, pyogenic granuloma, mucocele, benign salivary gland, or mesenchymal tumor, and lipoma were included in the differential diagnosis.

Radiographic examination revealed a faint welldefined circular with irregular radiopacity measuring 2 to 3 mm between 42 and 43 superimposed on the underlying normal bone architecture (Fig. 3). Routine hematological investigations were uneventful. The nodular growth and fibrotic growth were surgically excised. After taking a radiograph of the excised nodular gingival growth, both the specimens were sent for histopathological examination. Postoperative instructions were given and antibiotics were also prescribed. Healing was found to be satisfactory upon recalling the patient after 1 week and 1 month. The radiograph showed a well-defined circular radiopacity with irregular borders approximately 3 mm diameter surrounded by 1.5 mm radiolucent halo, suggesting POF (Fig. 4). The diagnosis was confirmed based on the histopathological report.



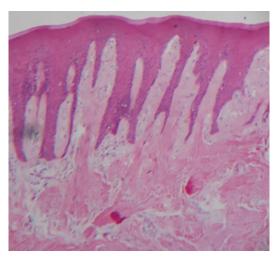


Fig. 5: Histopathological slide showing proliferating stratified squamous epithelium (4× magnification)

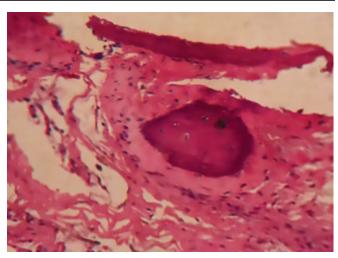


Fig. 6: The histopathological picture showing spongy bone (osteoid) (40× magnification)

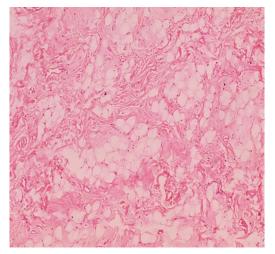


Fig. 7: Histopathological picture showing signet ring-shaped adipocytes with intervening areas of fibrosis (400× magnification)



Fig. 8: Follow-up image after 1 year with no recurrence

Histopathological report revealed:

- The section from attached gingiva showed covering of parakeratinized stratified squamous epithelium proliferating into underlying connective tissue with long and slender rete ridges. The subepithelial connective tissue was loose with moderate chronic inflammation and rest connective tissue being fibrous with dense bundles of collagen fibers and deeper areas showing ossification (Figs 5 and 6).
- The section from buccal mucosa showed a covering of a nonkeratinized stratified squamous epithelium spongiosus and proliferation into the connective tissue forming arcading pattern. The underlying connective tissue was fibrous with significant adipose tissue consisting of signet ring-shaped adipocytes seen (Fig. 7).

The histopathological report of the specimen was fibroepithelial hyperplasia with ossification and fibrolipoma respectively.

The patient came for a follow-up after 1 year and showed satisfactory healing with no recurrence (Fig. 8).

DISCUSSION

The name POF represents a misnomer as the term "fibroma" etymologically means tumor of fibrous connective tissue (from Latin fibra, "fiber" + Greek oma, "tumor"), but POF is not considered a true neoplasm.⁶ Intraoral ossifying fibromas have been described in the literature since the late 1940s. The POF was first reported by Shepherd in 1844 as alveolar exostosis. Eversole and Rovin⁸ later coined the term POF⁷ and stated that there were similar sex and site predilections along with similar clinical and histological features of pyogenic granuloma, PGCG or POF.⁸ It was also stated that these lesions simply vary in response to irritation. It has been suggested that POFs represent a separate clinical entity rather than a transitional form of pyogenic granuloma, PGCG, or irritation fibroma. The designation of peripheral

odontogenic fibroma according to the World Health Organization (WHO) has been given which reads that peripheral odontogenic fibroma is a rare and extraosseous counterpart of central odontogenic fibroma.⁹

The POF constitutes about 3.1% of all the oral tumors and about 9.6% of all the gingival lesions. 10 The POF is a reactive soft tissue growth that is usually seen on the interdental papilla and clinically appears as a solitary nodular mass, having a base that is either pedunculated or sessile. The high female predilection and a peak occurrence in the second decade and declining incidence after the third decade of life suggest hormonal influences, and POFs occur 2 to 4 times more frequently in females than in males between the ages of 25 and 35 years. The femaleto-male ratio reported in the literature varies from 1.22:111 to 1.7:1. 11-13 In the above-mentioned case, the female was 65 years. Approximately 60% of POFs occur in the maxilla and are found more often in the anterior region with 55 to 60% presenting in the incisor-cuspid region.¹⁴ In the presented case report also, the POF was pedunculated and present in the mandibular right anterior region in a female of 65 years.

Treatment requires correct surgical intervention which ensures deep excision of the lesion including the periosteum and affected PDL, which may reflect the technique and philosophy of surgical management. Thorough root scaling of the adjacent teeth and/or removal of other sources of irritants should be accomplished. Neville et al. Suggested that the lesion be removed down to the periosteum and the adjacent teeth be scaled to remove any remaining irritants. This will assist in lowering the rate of recurrence. In addition, POFs can cause erosion of bone, displace teeth, and interfere or delay eruption of teeth. The recurrence rate varies from 7 to 20% according to different authors. An important clinical aspect of POF is the high recurrence rate, which ranges from 8 to 45%. The POF shows a clinically benign behavior.

Various different surgical techniques, such as lateral sliding flap of full thickness or partial thickness, subepithelial connective tissue graft, or a coronally positioned flap, may be used to manage this defect and minimize esthetic patient concerns.

Our case presented with a rare coexistence of a fibrolipoma in right buccal mucosa.

Lipomas are rare soft tissue neoplasms in the oral cavity and account for 1 to 4% of oral benign tumors. ¹⁷ Oral lipoma was first described by Roux in 1848, who referred to it as "yellow epulis." ¹⁸ The etiology of lipomas is obscure, although mechanical, endocrine, and inflammatory causes have been attributed. ¹⁹ They are histologically classified into fibrolipoma, spindle cell lipoma, intramuscular or infiltrating lipoma, angiolipoma, sialolipoma, pleomorphic lipoma, myxoid, and atypical

lipoma. ¹⁶ Oral lipomas are slowly growing benign neoplasms presenting as a well-circumscribed, painless, submucosal nodule with a yellowish tinge. Oral lipomas are found commonly in males above 40 years of age. ²⁰ However, our case was a female patient aged 65 years. The buccal mucosa and the buccal vestibule are common sites where oral lipomas occur. ²¹

Fibrolipomas, classified as a variant of conventional lipoma by the WHO, occur commonly on the buccal mucosa and the buccal vestibule, followed by tongue, floor of mouth, and lips¹⁶ as seen in our case. Fibrolipoma differs from the classic variant because the mature adipose tissue is interspersed by bands of connective tissue.²² A recent study revealed that 27% of 41 cases of oral lipomas were fibrolipomas, whereas previous studies have reported a lower incidence. The treatment of oral lipomas and all histological variants is surgical excision. The prognosis of fibrolipoma is good and recurrence is rare.

CONCLUSION

This has been the first case report to show both POF in an uncommon location coexisting with fibrolipoma. Oral fibrolipomas are very rare in the oral cavity with few cases documented so far. Since the proliferative activity of fibrolipoma is greater than that of the other variants, the need for accurate diagnosis is important. The high recurrence rate reported for POF warrants the need for frequent follow-up regimen. More cases need to be reported to derive any significance for this coexistence.

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