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A Massive Peripheral Ossifying Fibroma–Uncommon Presentation of a Common Lesion

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Abstract

Peripheral Ossifying Fibroma (POF) is a relatively common gingival overgrowth whose pathogenesis remains uncertain. It predominantly affects adolescents & young adults mainly females, with a predilection for anterior maxilla. Clinically, it resembles a peripheral fibroma, but histopathologic analysis always reveals immature bone and osteoid within the lesion. The lesion is typically self-limiting and <2 cm,in size but there have been reports of uncommon huge peripheral ossifying fibromas. The mass-like clinical presentation and radiographic appearance of soft tissue densities may lead to misinterpretation of lesions larger than 2 cm, however the histologic appearance is diagnostic. Here we report an unusual case of POF in a 70-year-old female who presented with a massive localised gingival overgrowth in anterior mandible and a review of previously reported giant lesions.

Keywords: Peripheral ossifying fibroma, giant/massive peripheral ossifying fibroma, anterior mandible.

Introduction

Peripheral ossifying fibroma (POF) accounts for 3.1% of all oral tumors and for 9.6% of all gingival lesions[1,2]. Synonyms of POF are peripheral cementifying fibroma, calcifying or ossifying fibroid epulis, and peripheral fibroma with calcification[3]. In 1872, Menzel first described the ossifying fibroma, but only in 1927, Montgomery assigned its terminology[4]. The POF may appear ulcerated and erythematous or exhibit a color similar to the surrounding gingiva. It may be pedunculated or sessile and usually does not blanch upon palpation[5]. The POF may occur at any age, but exhibits a peak incidence between the second and third decades. The average age is around 28 years, with females being affected more often than males. Approximately 60% of POFs occur in the maxilla, often in the anterior than the posterior area, with 55%–60% presenting in the incisor-cuspid region. The lesion is typically self-limiting and <2 cm, however it has been recognized that some examples may grow quite large and may displace teeth[6]. The mass-like clinical presentation and radiographic appearance of soft tissue densities may lead to misinterpretation of

lesions larger than 2 cm, however the histologic appearance is diagnostic. The pathogenesis of this tumor is uncertain; however, the pluripotent cells of the periodontal ligament have the apparent ability to transform or metaplastically change into osteoblasts, cementoblasts or fibroblasts, in response to irritants such as calculus, bacterial plaque, orthodontic appliances, ill-adapted crowns and irregular restorations and are therefore, capable of producing a unique inflammatory hyperplasia, the peripheral ossifying fibroma[7,8,9]. Incidences of recurrence have been put at 16–20%[10]. As the clinical spectrum of this entity has resemblance to other common gingival masses, a thorough diagnostic work-up is necessary to rule out other common benign gingival lesions. We present a new case of massive POF and a review of previously reported giant lesions.

Case Report

A 70 year old female patient reported to the Department of Oral Medicine & Radiology, with the chief complaint of growth in the lower front gum region since 6 years. Her history of present illness revealed that patient noticed a small swelling in lower front region of the teeth 6 years back, which gradually increased to attain the present size. It was not associated with any pain or discharge but patient reported bleeding during brushing teeth. Patient did not give any history of trauma in relation to that region. The lesion was expansile and displaced the adjacent teeth. Concerning functional limitations, phonation, mastication and aesthetic, she stated that she had adapted to the mass so well she was able to eat to her satisfaction and speak relatively well and that she was not very concerned of the aesthetic appearance. So she had neglected the lesion and did not seek any treatment. The patient's medical history revealed that she was not currently under the care of any physician, had no known medical problems and was not currently taking any medications. No history of deleterious oral habits.

On clinical examination a solitary pedunculated well-defined exophytic lesion was present on the interdental gingiva in the region of 31 and 32[Figure 1&2]. It was spherical to oval in shape measuring approximately 6cm x 5cm x 4cm in size, pale in color. Multiple shallow erosive areas seen on anterior aspect with cresting at the centre. On palpation, it was slightly tender on the anterior aspect, where there was erosion, while in rest of the areas it was nontender. It was firm in consistency, mobile, noncompressible, nonreducible, and nonpulsatile. Patient had a very poor oral hygiene with an abundance of soft deposits and purulent exudates contributing to halitosis. Other findings were distally displaced 31,32 41, grade I mobile 31,41, and root remnant in relation to 11,22.

On the basis of history and clinical features, a provisional diagnosis of fibroma in relation to 31,32 was given. Clinically, the differential diagnosis of pyogenic granuloma, peripheral giant cell granuloma, peripheral odontogenic fibroma, and peripheral ossifying fibroma were considered.

Consequently patient was subjected for intra-oral periapical radiograph, complete haemogram, and excisional biopsy of the lesion. Routine haematological investigation values were found to be within normal limits. Intraoral periapical radiographic view showed distal displacement and periodontal ligament widening in relation to 31,32,41 and 42 and horizontal bone loss in relation to 31,32 and 41 [Figure 3]. The exicisional biopsy was performed under local anaesthesia. Histopathology showed [Figure 4] serial sections of single tissue bit composed of a para-keratinised stratified squamous epithelium overlying a moderate to densely collagenous stroma. Numerous calcifications are seen in the form of interconnecting bony

trabeculae, chiefly immature bone. Peripheral rimming by plump fibroblasts are noted around the trabeculae. Few areas of hyalinization are noted within stroma. Stroma is minimally infiltrated by chronic inflammatory cells, chiefly lymphocytes with moderate vascularity. Based on the patient's history, clinical, radiological and histological findings, the final diagnosis of peripheral ossifying fibroma (POF) with respect to 31,32 was given..

Discussion

Intraoral ossifying fibromas have been described in literature since the late 1940s. POF is usually solitary, rarely, it can be multicentric. Various names used for POF indicate that there is much controversy surrounding the nomenclature and classification of such lesions. Shepherd first reported this entity as “alveolar exostosis” in 1844. The term POF was coined by Eversole and Rovin in 1972 and Bhasker et al in 1984 described this lesion as peripheral fibroma with calcification[3,11]. Different terms have been used to describe this lesion like peripheral ossifying fibroma, peripheral cemento-ossifying fibroma, peripheral cementifying fibroma, peripheral fibroma with calcification, ossifying fibro-epithelial polyp, peripheral fibroma with cementogenesis, peripheral fibroma with osteogenesis, calcifying or ossifying fibrous epulis and calcifying fibroblastic granuloma which has been adding to confusion[11]. It is almost impossible to distinguish between ossifying and cementifying fibroma clinically and radiographically. The origins of POF are not clear. Some consider POF to develop secondary to fibrosis of granulation tissue because they resemble pyogenic granuloma clinically and histopathologically. Also, due to its predilection for female gender and second decade, the role of hormones has also been questioned. A widely acceptable histogenesis for POF is the inflammatory hyperplasia of the cells of the periosteum or periodontal ligament. Chronic irritation of the periosteal and periodontal membrane causes metaplasia of the connective tissue and result in initiation of formation of bone or dystrophic calcification. An origin from periodontal ligament is suggested because of exclusive occurrence of POF from interdental papilla and the proximity of gingiva to periodontal ligament. Other cited reasons include the presence of oxytalan fibres within the mineralized matrix of some lesions, the age distribution which is inversely related to the number of lost permanent teeth, and the fibro cellular response similar to other reactive lesions of periodontal ligament origin[11]. A case of multicentric POF at an edentulous site in a 49-year-old woman[12] has also been reported, further adding to the confusion regarding its etiopathogenesis. In the present case the local irritants might have been the cause of the growth.

POF presents as a pedunculated or sessile slow growing nodular mass with a smooth or ulcerated surface which may be pink to red in colour. The POF may occur at any age, but exhibits a peak incidence between the second and third decades. The average age is around 28 years, with females being affected more often than males. Eversole and Robin suggested that the loss of periodontium that accompany tooth loss in old age may explain its greater occurrence in the younger age group[3]. Approximately 60% of POFs occur in the maxilla, often in the anterior than the posterior area, with 55%–60% presenting in the incisor-cuspid region. However, in our case this lesion was seen in a 70 year old female in the anterior mandible, which makes it a rare entity. The lesion is typically self-limiting and <2 cm, however it has been recognized that some examples may grow quite large and may displace teeth[6]. Massive POF lesions are rare to find in clinical practice. For the purpose of the review on Giant POF,

only those measuring a minimum of 3 cm in at least 2 dimensions where considered. Giant POF turn out to be very uncommon, we were only able to find eighteen more cases in the world world literature in seventeen papers plus this case which accounts a total of nineteen.

Considering massive POF lesions in the literature [Table 1][13-30] all cases except six were reported in female patients with an average age of 50.61 years with age ranging from 10-70 years. The average size (largest dimension) was 6.08 cm with range from 3 to 10 cm. The average evolution time was 4.26 years with range between 2 months to 10 years. It would be interesting to study the molecular basis of such lesions to know the reasons for their enormous growth. The radiographic features of POF may range from no change to destructive changes depending on the duration of the lesion. In certain cases, superficial erosion of underlying bone, cupping defect and focal areas of radiopaque calcifications at the center of the lesion can be seen. Additional imaging studies are rarely required. However, if performed, Computed Tomography (CT) reveals it as a well circumscribed mass with evidence of calcification and mild enhancement after contrast agent administration. In Magnetic Resonance (MR) imaging, an isointense signal to muscle on non-enhanced T1 weighted sequence and an iso-to-low signal on T2 weighted sequence can be seen31. However, in the present case, no special radiographic imaging techniques were used. Histopathology provides the confirmatory diagnosis with the identification of fibrous connective tissue and the focal presence of bone or other calcifications as was seen in this case. Treatment of POF consists of elimination of etiological factors, scaling of adjacent teeth and total aggressive surgical excision along with involved periodontal ligament and periosteum to minimize the possibility of recurrence Long term post-operative follow up is extremely important because of the high growth potential of incompletely removed lesion and a relatively high recurrence rate of approximately 20%[10]. The average time interval for first recurrence is 12 months[32]. POF clinically resembles as pyogenic granuloma, peripheral giant cell granuloma or odontogenic tumors, so radiographic and histopathological examination is essential for accurate diagnosis.

Figures and Tables

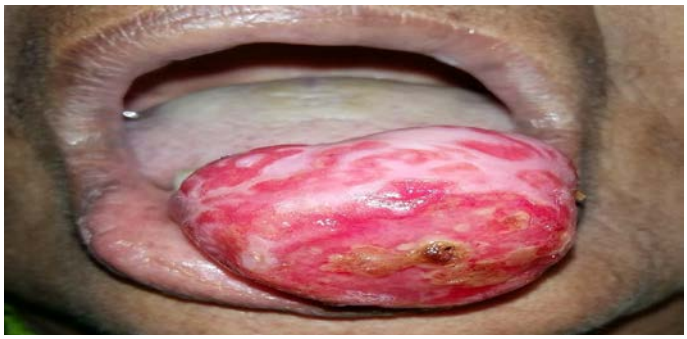

Figure 1: Facial view of the lesion

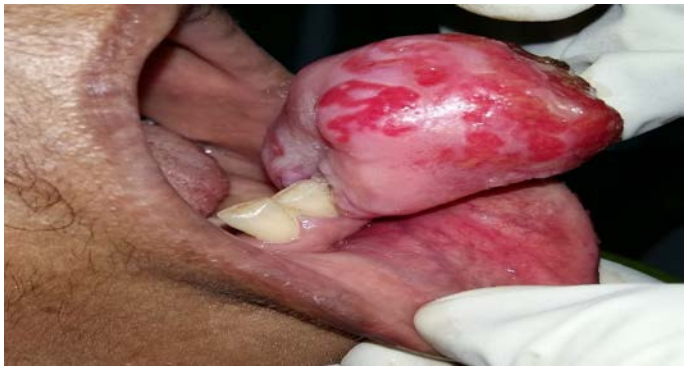

Figure 2 : Lateral view of the lesion

Figure 3]: Intraoral periapical radiograph showing distal displacement, horizontal bone loss and periodontal ligament widening in relation to 31,32 and 41.

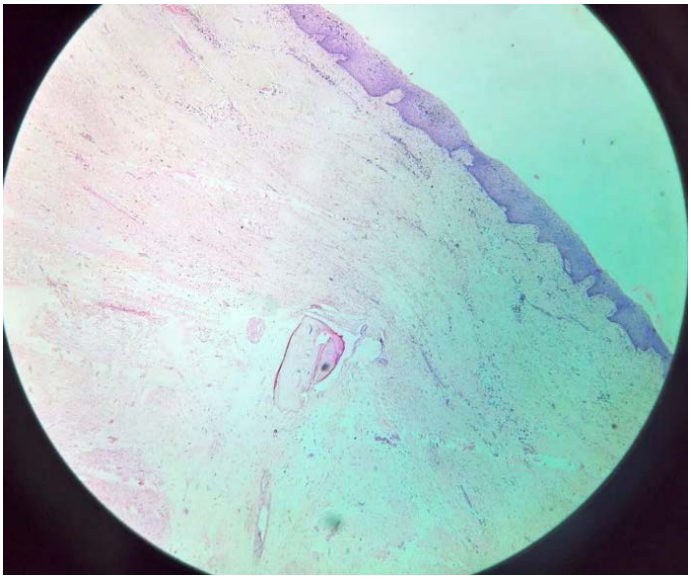

Figure - A

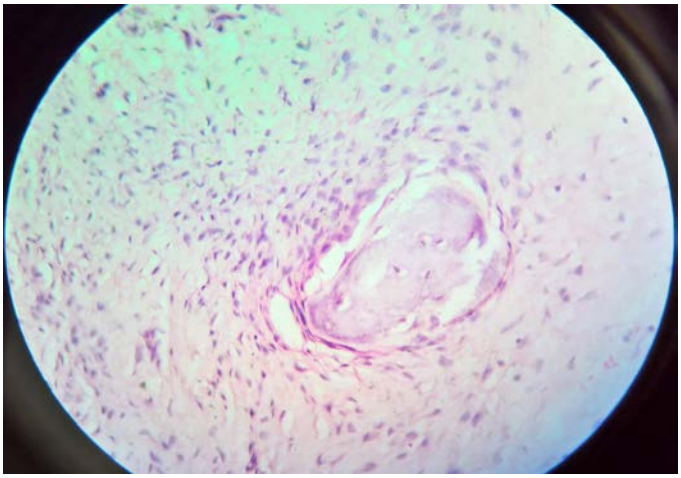

Figure – B

Figure 4: (A)H & E stained section showing overlying epithelium, fibrocellular connective tissue stroma and few eosinophilic calcifications (x40 magnification). (B) H & E stained sections showing calcification resembling bone. (x100 magnification).

Table 1

Author	**Patient Age/sex**	**Clinical features**	**Duration**	**Size**
Bodner and Dayan[13]	70 years/female	Location- Posterior mandible Asymptomatic. Pedunculated. Pink similar to mucosa.	5years	6cm
Poon et al.[14]	32 years/female	Location-Anterior maxilla Asymptomatic. Pedunculated.	5 years	9 cm

		Firm-rubbery in consistency		
Charro et al.[15]	68 years/female	Location- Posterior maxilla Asymptomatic. Pedunculated. Pink similar to mucosa.	10 years	5cm×5cm
Martins et al.[16]	32 years/female	Location- anterior maxilla Asymptomatic. Pedunculated. Pale pink Ulcerated Tooth displacement	5 years	5cm×4.5cm
Kim and Kim.[17]	66 years/female	Location-Posterior mandible Asymptomatic. Pedunculated. Pinkish,erythematous in ulcerated area Firm	5 years	8cm × 5 cm
Singh et al.[18]	70 years/female	Location- Anterior maxilla Asymptomatic. Pedunculated. Pink similar to mucosa Erythematous in ulcerated area Firm. Tooth mobility. Presence of abundant local irritants.	6 years	3 cm×3 cm
Vivekanandh et al.[19]	45 years/female	Location- Anterior maxilla Asymptomatic. Pedunculated. Pale pink with pigmentation. Smooth and firm	1 year	6cm ×7cm
Chaudhari and Umarji[20]	55years/female	Location-Posterior mandible Asymptomatic. Pedunculated. Pink similar to mucosa Firm	6 months	5.9cm
Trasad et al[21]	10years/male	Location- Posterior maxilla Asymptomatic. Pedunculated. Pink similar to mucosa.	3 months	6cm×3cm
Sacks et al[22]	52year/female	Location-Posterior mandible Asymptomatic. Pedunculated. Pink similar to mucosa Firm	1year	10.5cm
Manuel et al.[23]	64years/female	Location- Posterior maxilla	5 years	6cm ×7cm

		Asymptomatic Pedunculated(bilobed) Caused facial disfigurement No ulceration or bleeding		
Childers et al[24]	54years/male	Location- Anterior mandible Asymptomatic. Pedunculated Pink similar to mucosa Mostly Smooth surface Firm consistency. Tooth displacement and mobility	6years	4.5cm×3cm×3 cm
Grimaldo-Carjevschi et al.[25]	28years/female	Location- Anterior mandible Asymptomatic. Pedunculated. Pink similar to mucosa Mostly Smooth surface Hard consistency. Tooth displacement and mobility	14 months	5.3cm×4.5cm ×3.2cm
Himanshu and Ritika[26]	58 years/male	Location- Posterior mandible Asymptomatic PedunculatedReddish pink Smooth Smoker patient	2-3 months	5.5cm × 3cm × 2cm
Jitender Batra et al[27]	25years/female	Location- Posterior mandible Asymptomatic. Sessile. Pink similar to mucosa Mostly Smooth surface Hard consistency. Tooth displacement and mobility	8 months	4cm x 3.7cm
Reena et al[28]	62years/male	Location- Left maxilla and mandible Asymptomatic Sessile. Reddish pink Irregular lobulated surface Firm consistency. Tooth displacement and mobility	5 years	10cm x 6cm
Ronaldo et al[29]	38years/male	Location- Anterior mandible Asymptomatic. Sessile. Pink similar to mucosa Mostly Smooth surface Firm consistency. Tooth displacement and mobility	10years	3.5cm x 3cm

Vandana Reddy et al[30]	55years/male	Location- Posterior mandible Asymptomatic Pedunculated(bilobed) Caused facial disfigurement No ulceration or bleeding	6months	5cm×4cm
Present case	70years/female	Location- Anterior mandible Asymptomatic Pedunculated Pink+erythematous areas Mostly Smooth surface Firm consistency Tooth displacement and mobility	6 years	6cm × 5cm ×4cm

Conclusion

Massive peripheral ossifying fibroma have mainly been reported in females, with a probable mix of relation between growth factors and hormones. These lesions are uncommon even if under reporting is considered. Many factors must take part in the growth of these peripheral ossifying fibroma; and hence it would be of great interest to study the molecular basis of these lesions, with emphasis on growth factors. A socio-psychological reflection must also be considered, discussing why patients would allow these or other lesions to grow to the extent before looking for assistance.

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