

CASE REPORT

GIANT PERIPHERAL OSSIFYING FIBROMA OF THE MANDIBLE: CASE REPORT OF A RARE AND DISTINCT PATHOLOGY

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Received: 05 Feb, 2022

Accepted: 03 Mar, 2022

Published: 15 Mar, 2022

Key words: Clinical review; Giant peripheral ossifying fibroma; Gingival neoplasm; Oral reactive lesions.

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DOI: <https://doi.org/10.54530/jcmc.647>

Citation

Singh AK, Khanal N. Giant peripheral ossifying fibroma of the mandible: case report of a rare and distinct pathology. Journal of Chitwan Medical College. 2022;12(39):129-32.



Peer Reviewed

ABSTRACT

Peripheral ossifying fibroma usually presents as a small, pedunculated, gingival nodule, the pathogenesis of which is believed to be reactive rather than neoplastic. The lesion is typically less than two cm in size; however, there are reported cases where the lesion grew quite large. The oversize mass-like clinical appearance might be confused for other similar neoplastic lesions of the oral cavity, and the radiographic presentation of soft tissue calcification may lead to misdiagnosis; however, the histological features are diagnostic. There are very few cases reported of a giant POF; hence, we present a new case of a giant mandibular POF that was managed with resection and reconstruction. A brief review of previously reported cases on the clinical presentation, radiographic features, treatment modalities, and outcomes is performed to inform clinical decision making. This lesion represents a distinct clinical entity with a set of features requiring recognition for accurate diagnosis and a radical management approach due to its aggressive growth and a high recurrence rate.

INTRODUCTION

Peripheral ossifying fibroma (POF) is usually a small gingival nodule, the pathogenesis of which is reactive rather than neoplastic process.¹ The most common presentation is a pedunculated or sessile nodule attached to the gingiva and is derived from the periodontal ligament.² Young women are affected in a disproportionately high ratio, in their second and third decade, and the anterior mandible is the most common site of occurrence.²

The lesion is typically smaller than two cm and is grouped along with other reactive lesions of the oral cavity along with epulis, giant cell granuloma, gingival fibroma, and reactive giant cell lesions.³ However, there are few reported cases of giant mass-like lesions with the destruction of alveolar bone and displacement of teeth, which can readily be misdiagnosed as other common neoplastic lesions of the oral cavity. The atypical clinical presentation and radiographic appearance of soft tissue densities along with the displacement of teeth and massive destruction of the adjacent alveolar bone may lead to misinterpretation.⁴

Although there are large case series on hyperplastic fibrous

lesions of the periodontal origin, a substantial proportion of them include lesions smaller than two cm, and very few giant POF cases form the body of evidence to inform the clinical decision making required to recognize and manage these atypically large and aggressive lesions. This lesion represents a distinct clinical entity with a set of features requiring recognition for accurate diagnosis and a radical management approach due to its aggressive growth and a high recurrence rate.⁴

We present a new case of a giant mandibular POF with the focus on the clinical presentation, diagnostic pathway, radiographic features, histopathological features, treatment options, and the outcomes to inform the clinical decision making. The written consent was taken from the patient earlier for the preparation of this manuscript.

CASE PRESENTATION

History

A 24-year-old Asian female reported to the Oral and Maxillofacial clinic with a chief complaint of a large mass in her oral cavity that interfered with her speech and feeding.

The patient reported having experienced no pain in the area. The patient reported that the lesion began two years ago, which continued to grow and has reached the current size. The patient was of lower socioeconomic background, and her oral hygiene was poor. The medical history of the patient revealed that she had no known medical problems, and hence, currently not under any medications. Her vitals were within the normal range for her age, and all the lab parameters returned normal.

Clinical examination

Her extraoral examination was distinct, with a large lesion of approximately 9x5 cm visible on the left mandible. Rest of the facial bones were normal. Her mouth opening was also normal, but due to the lesion's large size, her oral aperture was blocked (Fig. 1A, 1B).

Intraoral examination revealed a non-tender, lobulated, pedunculated mass, extending from midline to the angle of left mandible, approximately 9x5 cm in size, covered by normal mucosa, non-ulcerated, with severe displacement and mobility of teeth from right second premolar to the left third molar. The mass was a little mobile, firm on palpation, extending from the alveolar mucosa, completely encroaching the mandibular corpus and further encroached upon the mandibular lingual plate. The patient had multiple caries, generalized advanced periodontitis along with halitosis. The large intraoral mass caused a lateral and posterior displacement of the tongue, and her speech was unintelligible. This mass effect of a large pedunculated gingival lesion displacing the tongue and affecting the patient's speech and feeding is a unique and very distressing presentation, not reported in the literature about a POF. The diagnostic considerations with such a presentation vary from benign aggressive odontogenic pathologies like ameloblastoma and central ossifying fibroma to malignant processes of mandible like osteosarcoma and primary central squamous cell carcinoma.



Figure 1: Clinical photographs A Frontal view of lesion of approximately 9x5 cm with normal overlying mucosa. B. Profile view of swelling in the left side.

Radiographic assessment

Orthopantomogram (OPG) revealed a poorly defined lesion with the appearance of diffuse soft tissue mixed opacity in the left mandibular body and displacement of all the ipsilateral mandibular teeth, though, There was no root resorption of the displaced teeth (Fig. 2). The left hemimandible was resorbed down to the inferior border from the dental midline to the angle of the mandible. CT scan revealed features consistent with calcifications and focal bone resorption as seen in ossifying fibromas.⁵

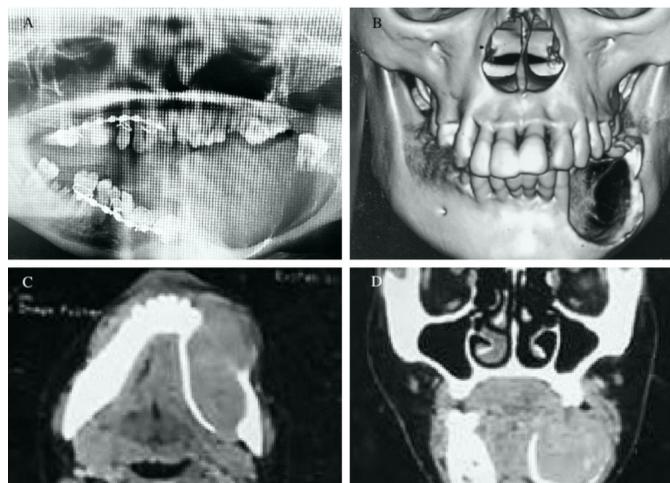


Figure 2: Radiographic Picture A. Orthopantomogram showing poorly defined lesion with mixed opacity in left side displacing the third molar. B 3D reconstruction view showing bony resorption in the left side C. Axial section with isodense lesion with hyperdense foci destroying both buccal and lingual cortical plate. D Calcification foci present in the coronal section.

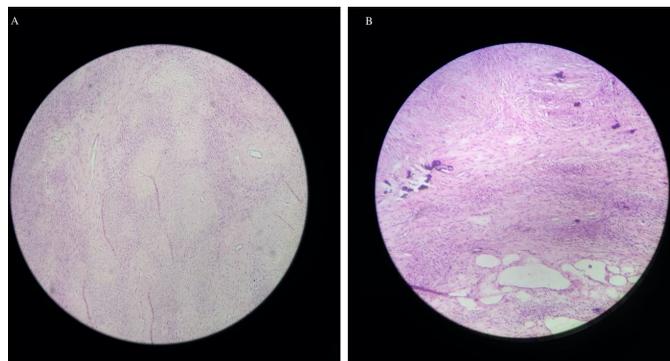


Figure 3: Serological Profile of Dengue Fever Patients

Histopathology

An incisional biopsy was performed and the specimen was sent for histopathological examination where the diagnosis of the lesion as POF was established. The calcified material exhibited a mixture of lamellar and woven bone when viewed under polarized light. Microscopic analysis of the Haematoxylin & Eosin-stained section revealed whorls and nests of benign spindle cells admixed with homogenous collagenous fibrous areas (Fig. 3). The fibrous component was cellular and predominantly comprised of plump fibroblasts. In

the connective tissue, few blood vessels were seen, and also revealed areas of calcifications & dark staining fragments of woven bone. Few myxoid areas were also seen.

Surgical management and follow-up

Due to the large and aggressive nature of the lesion and the known recurrence rate, we decided to manage the case with complete excision of the lesion along with the segmental resection of the mandible. The mandibular reconstruction was performed with a non-vascular block-graft from the Iliac Crest (Fig. 4).

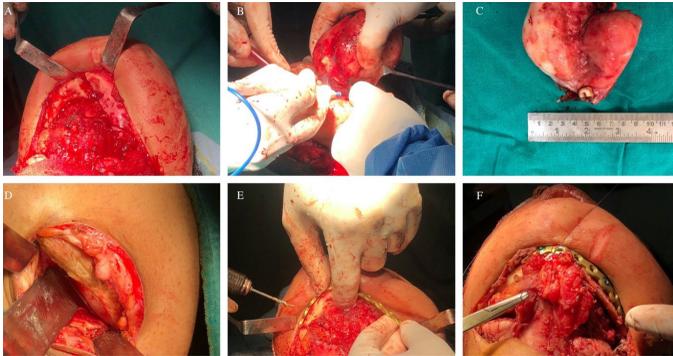


Figure 4: A: bilateral submandibular incision giving way to lower border of the mandible for resection. B: excision of the lesion after freeing the lesion from oral mucosa. C: excised lesion with measurement. D: exposure of the iliac bone for reconstruction E Use of reconstruction plate and fixation of iliac bone F Suspending the anterior belly of digastric to the reconstruction plate.

There was an immediate improvement in the patient’s speech and feeding, and the immediate postoperative period was free of any complications. There is no evidence of recurrence at one-year follow-up and we have planned for dental implant-supported rehabilitation after the consolidation of the bone graft (Fig. 5).



Figure 5: A: Facial view of the patient after surgery. B: intraoral showing normal, well-healed overlying mucosa. C: OPG showing reconstruction plate and iliac crest bone graft. D: 3D reconstruction CT scan showing a 2.5 mm locking stress-bearing plate holding the iliac crest bone graft.

DISCUSSION

The pathology was first described in 1844 by Shepherd et al. as alveolar exocytosis, and later, Eversol and Robin proposed the term POF in 1972.⁶ It is estimated that, POF represents 2–9 % of all gingival lesions amongst entire reactive lesions in the oral cavity, and additionally, after pyogenic granuloma and giant cell central granuloma, POF is the third most common lesion of all localized reactive hyperplastic lesions.³ The inflammatory reaction in POF is secondary to local irritants such as plaque and dental restorations. The chronic irritation of the periodontal ligament and the periosteum results in metaplasia of the connective tissue, that leads towards bone formation and hence, dystrophic calcification. The origin is from inflammatory hyperplasia in the cells of the periodontal ligament, the theory of which is fairly more accepted as peripheral ossifying fibroma seems to occur exclusively in the gingiva with close proximity to the periodontal ligament. The expression of osteopontin in all the cases of POF speculates that the origin is from the periodontal ligament for most of the cases of peripheral ossifying fibroma.⁷ The modality of treatment for peripheral ossifying fibroma ought to vary from other focal reactive proliferation of gingiva. Osterix protein has been observed to be significantly expressed in POF and may play a role in the pathogenesis of POF.⁸

Formerly reported data defined that two-thirds of the peripheral ossifying fibroma is seen in women, chiefly in the second decade of life, which may be related with the hormonal influences, particularly with progesterone and oestrogen as differences in their level during puberty and pregnancy can be reflected in the oral cavity with implications in the occurrence of gingivitis and fluctuations in crevicular fluid production, and might be involved in a higher incidence of POF during the second decade of life.² A contrasting finding was revealed in one immunohistochemical study in which the proliferating cells showed myofibroblastic characteristics without expressing oestrogen or progesterone receptors, which indicate that POF should be considered as a myofibroblastic proliferation, and despite the fact that the clinical characteristics suggest hormonal influence, the expression of hormone receptors in the proliferating cellular component could not be established.⁹ A higher incidence in the posterior region of the maxilla was reported in a case series by Hung et al. and an increased incidence (70.3 %) of POF in the mandible was described by Ojo et al. in 2014 ; in both the cases, variances in race and environment may have accounted for the variation.

There are very few cases of a giant variety of POF reported in the literature.⁴ Our case is 12th reported case of a Giant POF. These giant lesions are aggressive in growth, and cause destruction of the alveolar bone as well as displace and exfoliate adjacent teeth, as seen in our case. The microscopic characteristics consist of the presence of fibrous connective tissue with abundant fibroblast, collagen, profuse endothelial proliferation, and mineralized material. Treatment for the common variant of a small POF involves surgical excision of the lesion, including the periodontal ligament, periosteum, and tooth if they are affected; granting they have a good prognosis, the recurrence rate is

estimated to be 8–20 %, largely because of incomplete removal of the lesion. A new nomenclature for such large POF lesions has been recommended as Giant Peripheral Ossifying Fibroma (GPOF), as they differ significantly from smaller POF lesions and our case supports this cause.⁴ These Giant variants of POF is best treated by complete resection of the involved part of the mandible, usually segmental resection and reconstruction with a bone graft, followed by dental rehabilitation. For giant POF, if it is mainly located in the jaws without invasion of the skull base and/or pterygoid process and larger than 5 cm size radical surgical treatment should be performed for the prevention of tumour recurrence.¹⁰

CONCLUSION

The clinical case presented here is consistent with the common demographic characteristics of presentation. The localization of the lesion was frequently reported in the anterior maxilla, although mandible has also been documented, especially the giant lesions. The risk for recurrence is mostly associated with incomplete excision of the lesion. Recent information about this giant variant of POF is scarce, and an aggressive treatment plan is advocated for complete resolution of the condition.

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