Case Report

Unusually large-sized peripheral ossifying fibroma

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ABSTRACT

Fibrous growths in the gingiva with the histopathological presence of calcifications are a common occurrence in the oral cavity. These lesions can be neoplastic in nature with either odontogenic or non odontogenic origin or they can be reactive lesions. This is a case report of an unusual presentation of peripheral ossifying fibroma , unusual because of its abnormally large size with review of literature.

Keywords: Fibro-osseous lesion, maxilla, giant peripheral ossifying fibroma, reactive lesion

INTRODUCTION

The term ossifying fibroma has been used since 1927, and since 1968, cementum containing tumors has been grouped together. In 1971, the World Health Organization (WHO) classified cementum containing lesions into four types that include fibrous dysplasia, ossifying fibroma, cementifying fibroma, and cemento-ossifying fibroma.[1] However, the term "cementifying ossifying fibroma" was reduced to ossifying fibroma in the new WHO classification in 2005.[2] Ossifying fibromas are broadly classified as peripheral and central type. Peripheral ossifying fibroma is a localized reactive lesion, which is nonneoplastic and usually restricted to the soft tissues. In this case report, we present the clinical, radiological, and histopathological features of peripheral ossifying fibroma of an abnormally large size.

CASE REPORT

A male patient aged 62 years reported to our center with the chief complaint of dull gnawing pain and a growth in the mouth [Figure 1] of 5 years duration associated with difficulty in eating and swallowing. The history revealed that the patient had met with a road traffic accident 5 years back which resulted in injuries to his upper front teeth, lower lip, and left side of the face. A few months after the injury, he noticed a small growth on the left side of the upper gums that progressively increased slowly to the present size. There was a sudden surge in growth 20 days before presentation to our department after the patient removed a tooth fragment sticking to the growth. There was a gross asymmetry of face caused by the diffuse swelling involving the lower two-third of the left side of the face. Superoinferiorly, it extended from the infraorbital region to the lower border of the mandible and the anteroposterior extent was from the midline to the left preauricular region. The mass was reddish pale, exophytic with a lobulated surface, measuring approximately 10 cm × 6 cm and was protruding from the oral cavity causing inability to close the mouth. The growth was attached to the left maxillary alveolus and palate and not fixed to the buccal mucosa or vestibular sulcus. It did not show any focus of surface ulceration, but bleeding spots were distributed randomly. The upper posterior teeth were displaced and attached to the mass on the palatal side. On palpation, the growth was firm and tender with an irregular surface and was fixed to the upper left alveolus. A computed tomography angiogram was taken to check the vascularity of

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the lesion which revealed no irregular feeder vessels. Further imaging showed a soft-tissue shadow with calcifications. The soft-tissue shadow measured approximately 10 cm × 6 cm in size not involving the mandible but causing destruction of maxilla in the premolar region. Routine blood investigations and serology tests were done. Since it was an abnormally extensive lesion, we approached by giving Weber Fergusson with lateral limb modification connected with intraoral vestibular degloving incision to have a complete access of the lesion [Figure 2]. Then, peripheral osteotomy cut was given to facilitate easy release and that the lesion was excised completely out of the maxilla. The raw area was packed with iodoform gauze. Obturator was constructed postoperatively to close the palatal fistula. During follow-up, wound shrunk and obturator was replaced with maxillary prosthesis. The mass was sent for histopathological examination [Figure 3]. The weight of the excised mass was approximately 230 g. The differential diagnosis given for the mass was fibroma, ossifying fibroma, peripheral giant cell granuloma, pyogenic granuloma, fibrosarcoma. The histopathological examination of the soft-tissue specimen revealed fibrous, connective tissue with numerous plump fibroblasts [Figure 4]. Numerous areas showed ossification in the connective tissue. Chronic inflammatory cells and blood vessels were also seen. All of which suggested the mass be a peripheral ossifying fibroma [Figure 5]. Postsurgical recovery was uneventful and the 6 months follow-up revealed no evidence of recurrence.

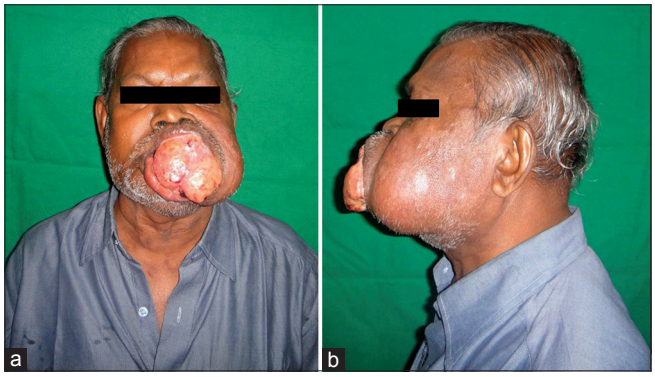

Figure 1: (a) Preoperative front view of the patient. (b) Preoperative profile view of the patient

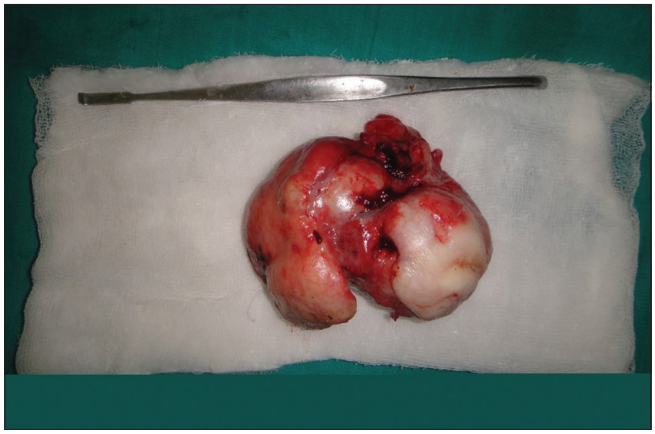

Figure 3: Complete excised lesion

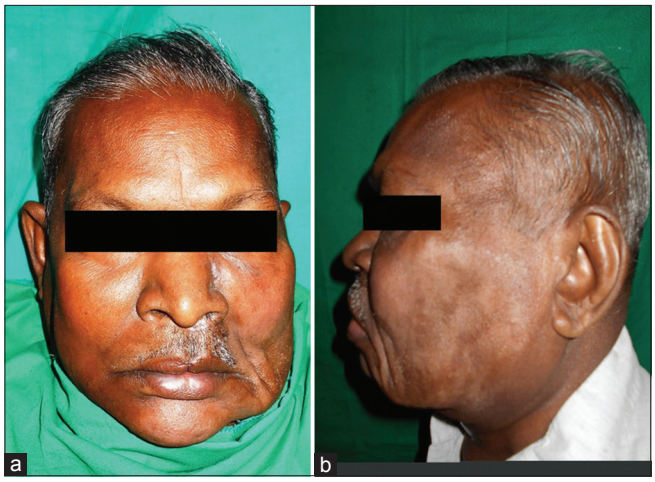

Figure 5: (a) Postoperative front view of the patient. (b) Postoperative profile view of the patient

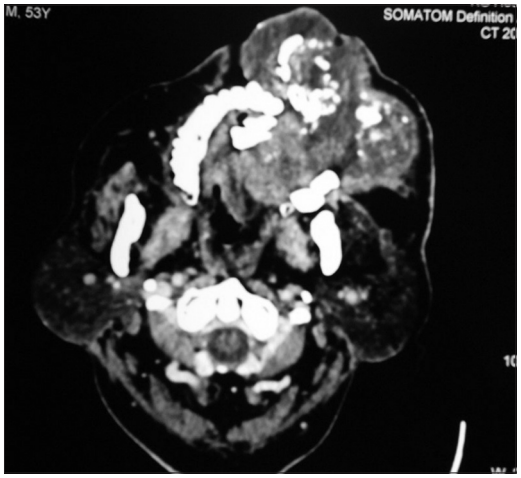

Figure 2: Computed tomography sectional view of the patient

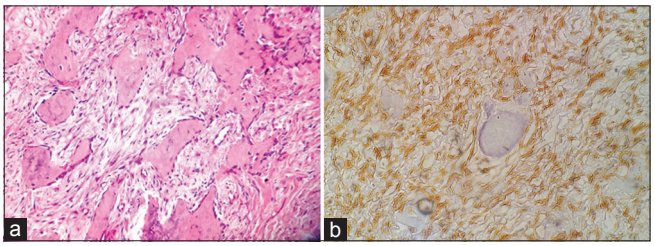

Figure 4: (a and b) Histopathological section of the excised lesion

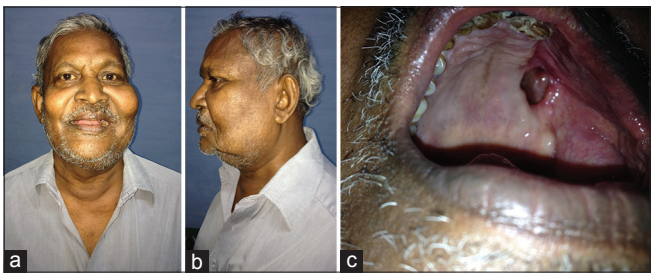

Figure 6: (a) Front view of the patient during follow-up. (b) Profile view of the patient during follow-up. (c) Intraoral view of the patient during follow-up

DISCUSSION

Fibro-osseous lesions of the jaws have been classified by Waldron and Giansanti into three distinct groups; (a) fibrous dysplasia, (b) fibro-osseous lesions arising in the periodontal ligament which would include central ossifying fibroma and peripheral ossifying fibroma, (c) fibro-osseous neoplasms of uncertain or debatable relationships, in which osteoblastoma and cementoblastoma have been categorized.[3]

Varying presentations of focal overgrowths have been manifested in the oral cavity containing different components of the connective tissue including collagen fibers, bone, cementum, and blood vessels. These focal overgrowths include pyogenic granulomas, fibromas, and peripheral giant cell granuloma. In the year 1872, Menzel described a series of lesions resembling ossifying fibroma, but Montgomery coined the term "ossifying fibroma" in 1927.[4,4a] In 1982, Gardner coined the term, "peripheral ossifying fibroma" for a lesion that is reactive in nature, and it is not the extraosseous counterpart of a central ossifying fibroma of the maxilla and mandible.[5]

Confusion existed with regard to the nomenclature of peripheral ossifying fibroma. Synonyms such as peripheral cementifying fibroma, ossifying fibroepithelial polyp, peripheral fibroma with osteogenesis, peripheral fibroma with cementogenesis, peripheral fibroma with calcification, calcifying or ossifying fibrous epulis, and calcifying fibroblastic granuloma. When bone predominates, "ossifying" is the term used, while "cementifying" is used when curvilinear trabeculae or spheroidal calcifications are encountered. When bone and cementum-like tissues are observed, the lesion is called "cemento-ossifying fibroma."[6]

Two types of ossifying fibroma have been cited in the literature, the central and the peripheral.[4,4b] Peripheral ossifying fibroma is a reactive lesion accounting for about 2% of all oral lesions that are biopsied. Eversol and Robin were the first to describe the lesion "peripheral ossifying fibroma" as a relatively uncommon, solitary, nonneoplastic gingival growth.[4,4c] Ossifying and cementifying fibromas are benign fibro-osseous lesions of periodontal ligament origin and therefore are histologic variations of the same neoplastic process. The lesion shows a sex predilection toward female and most commonly occurs in the second and third decades. Most commonly, they found in the maxilla and anterior to the molars, but numerous variations have been reported in literature.[7]

The etiology and pathogenesis of peripheral ossifying fibroma are unknown though many have suggested that these lesions originate in the cells of the periodontal ligament for the following reasons: The lesion occurs exclusively in the gingival tissue, close to the periodontal ligament; oxytalan fibers are reported to be found within the mineralized matrix of some lesions; the age distribution of the lesion is inversely proportional to the number of permanent teeth lost; the fibrocellular response is similar to that of other reactive gingival lesions originating in the periodontal ligament.[8] Hence, local irritants such as dental plaque, calculus, microorganisms, masticatory forces, ill-fitting dentures, poor-quality restorations, and trauma have all been implicated in the etiology along with the suggestions of hormonal influence because of the high occurrence in females.[9]

The size of peripheral ossifying fibroma, as reported in the literature, ranges from 0.4 to 9.0 cm with an average lesion measuring approximately 1.0–2.5 cm at its greatest dimension.[8] One such case of giant cell ossifying fibroma of maxilla measuring about 9 cm was first reported by Poon *et al*. in the year 1995.[10] Childers *et al*. suggested a distinct subset giant peripheral ossifying fibroma for abnormally large reported cases of peripheral ossifying fibromas.[11] However, in this case, the lesion has reached a gigantiform size of 10 cm × 6 cm causing destruction of the maxilla adjacent to the lesion and stretching the cheek tissues due to the prolonged duration of the large lesion, which may have resulted in tissue histogenesis. In spite of the large size of the lesion, it was noted to be less infiltrative as reported in literature.

The confirmatory diagnosis of lesions of this kind is usually achieved by histopathological examination. Histologically, the key feature of this lesion is exceedingly cellular mass of connective tissue comprising large number of plump fibroblasts intermingled throughout with delicate fibrillar stroma. The following features are usually observed during the microscopic examination:[12] (1) Intact or ulcerated stratified squamous surface epithelium; (2) benign fibrous connective tissue with varying numbers of fibroblasts; (3) sparse to profuse endothelial proliferation; (4) mineralized material consisting of mature, lamellar or woven osteoid, cementum-like material, lamellar or dystrophic calcifications; (5) acute or chronic inflammatory cells in lesions.[13] All the above features were seen in our case, thereby confirming our diagnosis. Buchner and Hansen observed that the mineralized tissues in peripheral ossifying fibroma can be of three basic types. (a) Bone that may be woven, lamellar, or trabecular, sometimes surrounded by osteoid. (b) Cementum-like material that appears as spherical bodies or large acellular round to oval eosinophilic bodies which coalesce to form islands of various sizes and shapes. (c) Dystrophic calcification which can range from small clusters of minute basophilic granules or tiny globules to large solid irregular masses.[12]

Immunohistochemical profile studies indicate that the proliferating cells are of a myofibroblastic nature, i.e., cells sharing morphologic characteristics with fibroblasts and muscle cells. C68 positive histiocytic component intermingling with lymphocytes and plasma cells suggests the existence of a reactive phenomenon or a response to inflammation.[7]

Multicenter peripheral ossifying fibroma can occur in oral and maxillofacial regions in conditions associated with known genetic mutations such as Nevoid basal cell carcinoma syndrome, multiple endocrine neoplasia Type II, neurofibromatosis, and Gardner's syndrome.[14]

The treatment for this type of reactive lesion is usually complete surgical excision along with curettage of the adjacent tissues to prevent recurrence.[15,16] In this case, we approached by giving Weber Fergusson with lateral limb modification along with intraoral vestibular degloving incision to have complete access of the lesion. The recurrence rate is high which varies from 8% to 20% as these reactive lesions which may probably be attributed to incomplete removal, repeated injury, or persistence of local irritants. Lasers are being used for treating peripheral ossifying fibromas as this gives a bloodless field and also minimizes scarring

and wound contraction.[17] Mergoni *et al*. showed 30% recurrence among 27 cases of peripheral ossifying fibromas.[18] Taking into the consideration the size, duration, the potential for recurrence and also as we were yet to make sure of disease free, we opted to wait and followed up the patient periodically. Since then, we managed to close the defect with maxillary prosthesis and thereupon the patient was satisfied with that which enabled him to bring him back to function. The reconstruction options available for such maxillary defects could vary from simple prosthesis to using locoregional flaps such as temporalis muscle and fascia along with buccal fat pad or skin graft or to distant flaps such as anterolateral thigh free flap or free fibula flaps, followed by dental rehabilitation using implants.[19] Booth suggested smaller margins than the 1 cm margin that is required for ameloblastoma, odontogenic myxoma, or calcifying epithelial odontogenic tumor.[20]

Eversole and Leider in a study of 64 cases of central ossifying fibroma reported a recurrence rate of 28%, following surgical curettage of these lesions.[6] With peripheral ossifying fibromas, incomplete removal can result in recurrence. Our patient has been reviewed for 24 months, with no evidence of recurrence [Figure 6].

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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