Central Ossifying Fibroma of Mandible – A Case Report

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ABSTRACT

The term ossifying fibroma (OF) is a slow-growing benign neoplasm of bone that occurs most commonly in the jaw. OF was first described by Menzel in 1872. If the lesions are small, they are treated by enucleation. However, larger lesions require radical resection, but some lesions behave aggressively, reaching massive proliferation, thus demanding careful management and follow-up. Recurrence rates of these aggressive forms of ossifying fibromas are about 30–38%. OF requires radical surgery because of the tendency for recurrence and the possibility of malignant transformation. In the present case report, a 25 years old female patient diagnosed with OF who underwent marginal resection for the same experienced recurrence after resection. The presentation and management of the case, along with review of the literature were discussed in this paper.

Key words: Fibro-osseous tumor, marginal resection, ossifying fibroma

INTRODUCTION

Central ossifying fibroma (OF) is a part of a spectrum of fibro-osseous lesions of the jaws. They are rare, benign, non-aggressive tumors that are commonly seen in head and neck region. Fibro-osseous lesions are diverse group of processes that are characterized by replacement of normal bone by fibrous tissue containing a newly formed mineralized product.^[1] OF is a rare, destructive, deforming, slow-growing, and benign fibro-osseous tumor. It is usually found in the craniofacial bones, with the mandible being the most common site. Computed tomography (CT) imaging plays a major role in detecting the extent of such lesions, their diagnosis, and planning the management.

In 1968, Hamner *et al.* analyzed 249 cases of fibro-osseous jaw lesions of periodontal membrane origin and classified them.^[2] In 1973, Waldron and Giansanti reported 65 cases (of which 43 cases

Quick Response Code	Article Info:
回想を回	doi: 10.5866/2019.11.10131
1993 - 19 13	Received: 06-09-2019
line states of	Revised: 05-10-2019
	Accepted: 14-10-2019
	Available Online: 02-01-2020, (www.
	nacd.in) $\ensuremath{\mathbb{C}}$ NAD, 2020 - All rights reserved

had adequate clinical histories and radiographs) and concluded that this group of lesions was best considered as a spectrum of processes arising from cells in the periodontal ligament.^[3] In 1985, Eversole *et al.* described the radiographic characteristics of central OF, and two major patterns were noted, expansile unilocular radiolucencies and multilocular pattern.^[4]

CASE REPORT

A 25-year-old female patient reported to the Department of Oral and Maxillofacial Surgery, Kamineni Institute of Dental Science with a chief complaint of painless swelling in the left side of the mandible for 6 months. The patient had difficulty in speech and chewing due to the swelling. On extraoral examination, the face was apparently symmetrical, and there was no sign of palpable lymph nodes. Intraorally, a solitary swelling was seen on the anterior mandibular region the size of 2 2 cmapproximately, extending from left second premolar to right canine (It crossed the midline). Both lingual and buccal plate expansion were seen and lingual expansion was evident [Figure 1]. On palpation, swelling was smooth, non-tender, and bony hard in consistency. Mucosa over the lesion was normal.

CT showed expansile lytic lesion with patchy sclerosis measuring about 2 2 cm in size

(Transverse Anteroposteriorly) [Figure 2]. A tooth is also seen within the lesion. The central densities within the lesion were of soft-tissue density, suggesting solid or fibrous lesion rather than a cystic lesion. The CT revealed a OF, that due to patchy sclerosis. The incisional biopsy was done under local anesthesia (2% lignocaine with adrenaline (1:80000) and specimen was sent for histopathological examination.

Histopathological findings revealed that the connective tissue stroma is hyper cellular with evidence of eosinophilic ossifying areas showing lacunae containing osteocytes and peripheral osteoblastic riming. The connective stroma was fibro cellular containing numerous spindle-shaped fibroblasts and numerous small and large basophilic globular bodies. These features were suggestive of



Figure 1: Clinical picture showing the extension of the lesion from 35 to 42 regions



Figure 2: Computed tomography of the coronal section showing superoinferior extension of the lesion

ossifying fibroma. Then, the patient was planned for surgery under general anesthesia.

Anterior vestibular incision was given from 35 to 46, full-thickness mucoperiosteal flap was raised from 35 to 46 region reflection [Figure 3] and osteotomy cuts were marked from of 35 to 43, osteotomy was carried out followed by marginal resection of the segment [Figure 4]. Layer wise primary closure was done using 3-0 Vicryl. Pressure dressing given. One day post-operative orthopantomograms showed intact inferior border of the mandible [Figure 5].

The patient was recalled after 1 month for followup when a small swelling was seen in the defect site. Incisional biopsy was performed on the swelling and was sent for histopathological examination. Histopathology report indicated OF. CT was advised, followed by resection and reconstruction.



Figure 3: Clinical picture showing the exposure of the surgical site and the identification of the lesion



Figure 4: Resected pathological lesion

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Surgery was planned under GA. Right nasotracheal intubation was done uneventfully. About 2% lignocaine with adrenaline (1:80000) infiltration was given. The lower vestibular incision was given from 36 to 46, full-thickness mucoperiosteal flap was raised from 36 to 46 regions, reflection was done, buccal and lingual segmental resection was done in the region of 35–42. Eyelet loops are placed in molar region, and IMF was done, and occlusion was achieved.

2.5 mm RECON plate was placed from 36 to 43 regions and stabilized with 2.5 8 mm screws on adjacent to the defect area [Figure 6]. Genioglossus and geniohyoid muscles are sutured to the recon plate with 2-0 Prolene. Closure was done with 3-0 Vicryl and the patient was extubated and recovered



Figure 5: One-day postoperative orthopantomograms showing the intact border of the mandible



Figure 6: Segmental resection was done and the two bone segments were stabilized with recon plate

uneventfully with pressure dressing given. Followup of the patient was done regularly at 2 months, 4 months, and 6 months. The follow-up period revealed uneventful wound healing

DISCUSSION

In 1891, von Recklinghausen introduced the term osteitis fibrosa to describe those osseous lesions in which marrow had been replaced with fibrous connective tissue. OF was first described by Menzel in 1872. It is a rare, benign primary bone tumor that occurs most commonly in the jaw. Montgomery in 1927 coined the term "OF."^[2] When this tumor arises in children, it has been named the juvenile aggressive OF, which presents at an early age and is more aggressive clinically and more vascular on pathologic examination.^[3]

In 1971, the World Health Organization (WHO) classified four types of cementum-containing lesions: Fibrous dysplasia, OF, cementifying fibroma, and cemento-ossifyingfibroma.^[4] According to the second WHO classification, benign fibro-osseous lesions in the oral and maxillofacial regions were divided into two categories, osteogenic neoplasm and non-neoplastic bone lesions; cement-OF belonged to the former category.^[5] However, the term "cemento-ossifying fibroma" was reduced to OF in the new WHO classification in 2005.^[6]

The origin of ossifying fibroma is thought to be the periodontal membrane. Some OFs do, in fact, contain prevalent cementum such as calcifications and others show only bony material, but a mixture of the two types of calcification is commonly seen in a single lesion.^[7]

Waldon described the cementifying and OFs of the jaws as well-circumscribed, slow-growing, painless, expansile lesions that occur over a wide age range, with peak incidence during the third and fourth decades.^[8] The most common location is the premolar-molar region of the mandible. Radio graphically well-circumscribed, these lesions show varying degrees of radiolucency. Although adjacent teeth may be displaced, root resorption is not a feature of this lesion.

According to Fu and Perzin, considerable controversy and confusion still exist concerning some fibro-osseous lesions that involve the bones of the head, especially OF and fibrous dysplasia.^[9-12] OF has an osseous capsule, whereas fibrous dysplasia does not. The bone trabeculae are usually surrounded by osteoblasts and occasionally by osteoclasts.

Such components constitute an essential feature to differentiate this lesion from fibrous dysplasia.

Although MacDonald-Jankowski considered that radiological diagnoses were not difficult for specialist radiologists, not all radiological diagnoses in the reports were in accordance with the final histological diagnoses. The radiographic characteristics of the tumor have two basic patterns: Cystic lesion (unicystic or multicystic) and mixeddensity lesion. The radiographic borders of the tumor appear relatively smooth, well defined, and mostly corticated. The contour is regular.^[10]

The lesion tends to be concentric within the medullary part of the bone, with outward expansion approximately equal in all directions. This can result in the expansion of the outer cortical plate of bone. Although displaced and thinned, the lesion remains intact. The tumors always grow along the body of the jaws, even involving the whole jaw. This phenomenon suggests that OF may display aggressive local growth.

The treatment of choice for OF is surgical excision. Enucleation and curettage could be suitable for small and well-defined lesions; however, larger masses require radical surgery. The size of the lesion, level of cortical bone invasion, and involvement of the surrounding soft tissues are the main determining factors in the choice of treatment option.^[11] OF requires radical surgery because of the tendency for recurrence and the possibility of malignant transformation. All reported patients with partial or incomplete resection experienced the recurrence.^[12]

Most of the authors consider fibrous dysplasia and OF to be histologically similar – with the sole differentiating feature being that a fibrous capsule surrounding the latter is rarely observed in the case of fibrous dysplasia. However, an aggressive form of ossifying fibroma may lose its fibrous capsule.

If the OF lesions are small, they are treated by enucleation. However, larger lesions require radical resection. Recurrence rates of these aggressive forms of ossifying fibromas are about 30–38%. Liu *et al.* observed that the time of recurrence was always unpredictable, ranging from 6 months to 7 years after the operation. Hence, long-term follow-up of such cases is advisable for at least 10 years.^[13-16]

In the present case, a 25 years old patient came with the chief complaint of swelling in the left lower anterior mandibular region for 6 months. Routine investigations and incisional biopsy were done which were found to be OF. Based on the size and site of the lesion resection should have been done. However, considering the age and esthetics of the patient and nature of the lesion marginal mandibulectomy was planned for the patient. Routine follow-up 1 month postoperatively patient again presented with complain of swelling of size, 0.5 cm for which incisional biopsy performed, histopathological examination was again reported as OF. Later resection was done followed by reconstruction with titanium RECON plate. Followup was done 2, 4, and 6 months postoperatively and the patient showed satisfactory healing and no signs of recurrence.

CONCLUSION

The OF of the mandible is an uncommon benign tumor. Cosmetic, dental, and occlusal problems are often the first manifestations of these lesions, as they are clinically asymptomatic. CT imaging plays a major role in determining the extent of such lesions, their diagnosis, and treatment planning. Surgical curettage or enucleation with a long-term follow-up is the initial treatment of choice for small central OFs, whereas surgical resection is indicated for the large lesions. Resection is a better treatment plan in recurrent cases.

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