



Cemento-ossifying Fibroma of the Mandible: A Benign but Potentially Destructive Tumor

¹SK Kaushik, ²Shashidevi Haranal, ³Nitesh Naresh

ABSTRACT

Cemento-ossifying fibroma (COF) is a benign fibro-osseous tumor containing variable amount of mineralized tissue resembling bone or cementum. Microscopically it exhibits actively proliferating fibroblasts with varied cellular pleomorphism and mitotic activity. We present a case of a woman referred for evaluation of bony swelling in the lower half of the face. The mass had first appeared 3 years ago and was growing larger. The clinical, radiological, histopathology and the successful surgical management of the case is discussed here.

Keywords: Fibro-osseous, Tumor, Cementum.

How to cite this article: Kaushik SK, Haranal S, Naresh N. Cemento-ossifying Fibroma of the Mandible: A Benign but Potentially Destructive Tumor. *Oral Maxillofac Pathol J* 2015; 6(1):561-564.

Source of support: Nil

Conflict of interest: None

INTRODUCTION

Menzel gave the first description of a variant of ossifying fibroma, which was a benign fibro-osseous neoplasm, calling it as a cemento-ossifying fibroma (COF) in the year 1872.¹ In 1992, the World Health Organization (WHO) revised the nomenclature, and the separate lesions of cementifying fibroma and ossifying fibroma were named as a single entity of 'cement-ossifying fibroma'. COF bears close resemblance to lesions, such as fibrous dysplasia, cementifying periapical dysplasia or cemento-osseous florid dysplasia.²

It is believed to originate from cells of the periodontal ligament (PDL). PDL, layer of fibrous connective tissue surrounding the roots, contains multipotential cells that can form cementum, lamellar bone and fibrous tissue.

Under pathological conditions, neoplasms containing any or all of the components may be produced.^{3,4} COF can occur at any age but commonly during third and fourth decades of life and restricted to the tooth bearing areas of the jaws, often in premolar-molar region of the mandible.⁴

It is a progressively growing lesion that can attain an enormous size with resultant deformity if left untreated. We present a case of a wife of a serving soldier who was diagnosed and treated successfully for a large destructive COF.

CASE REPORT

A 35-year-old female patient came with a swelling on her right mandible of 6 months duration. There was facial deformity, moderate pain and difficulty in chewing. Clinically, there was a marked swelling measuring 5 × 7 cm on the right body of the mandible which was firm and nontender on palpation (Fig. 1). Intraorally, tumor caused a buccolingual expansion, obliteration of the vestibulum over the region of 45 to 47, slightly displacing tooth 45 (Fig. 2) and 46 was missing, extracted 3 years back. The mass was firm and not tender on palpation. The surface texture of the overlying mucosa was intact.

Panoramic X-ray showed round shape radiolucency on the left mandible extending from the distal aspect of 43 toward the region of 47. It has sharp and sclerotic margin, showing multiloculation and spots of radiopacity in the center of the lesion (Fig. 3). CT scans showed buccal and lingual cortical expansion (Fig. 4). Provisional diagnosis was considered as an ameloblastoma, calcifying epithelial odontogenic tumor or a fibro-osseous neoplasm. The incisional biopsy of the lesion revealed the microscopic diagnosis of COF.

The tumor was removed together with the adjacent bone with segmental resection of the right mandible (Fig. 5). The defect in the mandible was reconstructed by placement of bridging plate using stainless steel reconstruction plate. The histopathologic result shows that the tumor consists of fibroblast proliferation with foci of cementum-like material (Figs 6 and 7) which confirmed the diagnosis of COF.

A postoperative OPG was taken (Fig. 8) and the patient was reviewed during her continuous follow-up appointments and was also rehabilitated for the missing teeth (Fig. 9).

¹Group Captain (Classified Specialist), ²Reader
³Major (Graded Specialist)

¹Department of Oral and Maxillofacial Surgery; Commanding Officer, 8 Air Force Dental Center, Kanpur, Uttar Pradesh, India

²Department of Oral and Maxillofacial Surgery, Rama Dental College and Hospital, Kanpur, Uttar Pradesh, India

³Department of Oral and Maxillofacial Pathology, 316 Field Hospital, Indian Army, Rajasthan, India

Corresponding Author: Nitesh Naresh, Major (Graded Specialist), Department of Oral and Maxillofacial Pathology 316 Field Hospital, C/O 56 APO, Indian Army, Rajasthan, India Phone: 09657553199, e-mail: nitesh.naresh@gmail.com



Fig. 1: Patient in the case report showing swelling in the region of the lower right mandible



Fig. 2: Intraoral photograph showing buccolingual expansion over right posterior region



Fig. 3: OPG showing a well-defined radiolucency of the areas of foca radiopacities root displacement of 45, missing 46

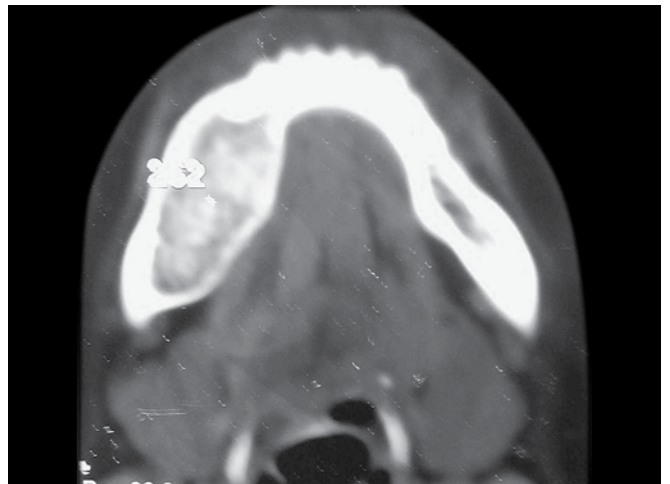


Fig. 4: Computed tomography scan showing expansion of the buccal and lingual cortices of the mandible



Fig. 5: Enucleated lesion of the patient mentioned in this case report

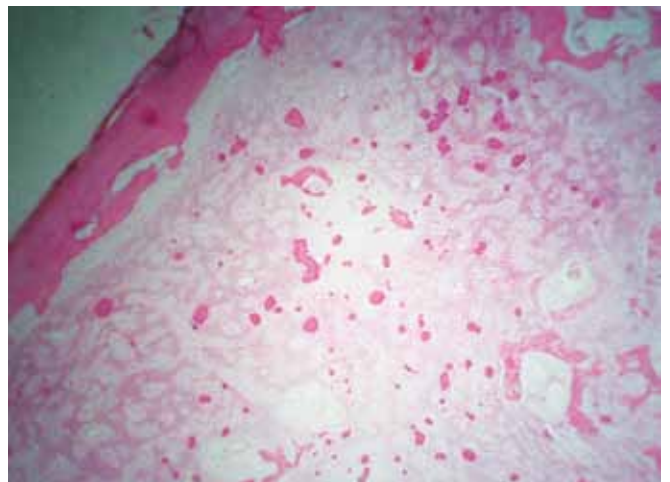


Fig. 6: Photomicrograph of the histopathological examination (H&E staining) of the lesion showing highly cellular fields with calcified areas (10x)

DISCUSSION

The concept of fibro-osseous lesions (FOL) of bone has evolved over the last several decades and now includes two major entities: fibrous dysplasia and ossifying

fibroma, as well as the other less common lesions, such as florid osseous dysplasia, periapical osseous dysplasia, focal sclerosing osteomyelitis, proliferative periostitis of Garre and osteitis deformans.⁵ In recent years, these

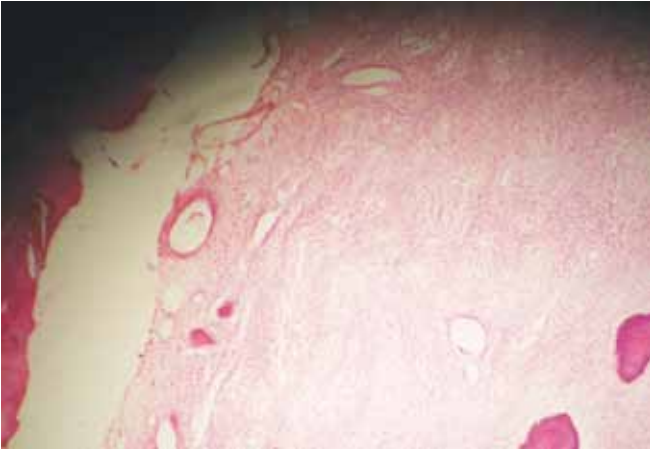


Fig. 7: Photomicrograph of the histopathological examination (H&E staining) of the lesion showing highly cellular fields with calcified areas (40x)



Fig. 8: Postoperative OPG showing the reconstruction plate

lesions were reclassified into fibrous dysplasia, reactive (dysplastic) lesions arising in the tooth-bearing area, and fibrous osseous neoplasms such as cementifying and ossifying or COF.^{6,7} Cementum is the mineralized connective tissue that covers the root of the teeth. The hybrid term, central COF is used because there is a variety of FOLs arising from the PDL, ranging from those with only deposition of cementum to those with only deposition of bone.⁸

Central COFs occur more frequently in women than in men. They arise in the mandible in 62 to 89% of patients, 77% occurring in the premolar region.⁸ Central COFs are asymptomatic until they cause expansion. Thus, they are generally not diagnosed until the tumor has had time to produce calcifications. In our case, the COF presented in a 35-year-old lady in the premolar-molar region and the patient also reported after the tumor had grown to a very large size causing a disfigurement.

Its exact etiology is still unknown, available data suggest either a reactive or developmental origin, majority of the cases in literature have been found to have a history of trauma in the area of the lesion. In accordance with the data found in the literature, our patient reported to have suffered from trauma in the affected area 8 years ago.⁹

Radiological features of COF can differentiate it from other FOLs. The lesions may be either unilocular or multilocular.¹⁰ In the early stages, COF appears as a radiolucent lesion with no evidence of internal radiopacities. As it matures, there is increasing calcification so that the radiolucent area becomes flecked with opacities until ultimately the lesion appears as an extremely radiopaque mass. COF presents a radiolucent appearance in 53%, a sclerotic radio density in 7% and mixed or mottled appearance in 40% of the cases.¹¹

The microscopic findings mirror the radiographic findings. The more radiolucent lesions are composed



Fig. 9: Prosthetic rehabilitation of the patient

of cellular fibrous connective tissue, frequently in a whorled pattern.¹⁰ Calcific deposits are seen throughout the fibrous stroma. Irregular trabeculae of woven bone or lamellar bone are most consistently noted in these tumors. Additional patterns of calcified material include small, ovoid to globular, basophilic deposits and anastomosing trabeculae of cementum-like material.¹² However, recognition of these structures is important in establishing its diagnosis.¹³ In our case, the histopathology was in accordance to the literature and showed rounded calcific, basophilic cementum-like deposits in the fibrous connective tissue.

Treatment of COF generally has been by conservative enucleation or curettage or radical surgery^{12,14} depending on the size and location of the individual lesion.¹⁵ Sloopweg and Muller¹⁶ reported that there was no difference in outcome between patients treated in a more limited way and those treated by major surgery. Other authors, however, advocate more extensive surgery for more aggressive lesions and lesions involving craniofacial bones in light of the potential for recurrence.^{17,18}

Sakoda et al¹⁹ described the procedure of a segmental resection of an extensive ossifying fibroma with the replacement of the excised segment with immediate reconstruction which we too have followed in our case.

In our opinion, the treatment was reasonable since the patient came with relatively large tumor and had history of rapid increase in size which might indicate increased aggressiveness of the lesion. Moreover, it is almost impossible to accomplish complete excision of the tumor in cases when the size of the tumor is extensively large only with surgical curettage through intraoral approach, not to mention the higher risk for mandible fracture following curettage procedure in large tumors.

An interdisciplinary approach is required to diagnose a COF from clinical, radiographic and microscopic features. These tumors may exhibit variations in their neoplastic behavior, it is therefore, important to take into account the individual tumor behavior when one is planning a proper surgical treatment, in order to eliminate the tumor completely and avoid tumor recurrence and at the same time improve the patient's cosmetic and functional problems.

REFERENCES

1. Hamner JE 3rd, Scofield HH, Cornyn J. Benign fibro-osseous jaw lesions of periodontal membrane origin: an analysis of 249 cases. *Cancer* 1968;22:861-878.
2. Ghom AG. Text book of oral medicine. 2nd ed. New Delhi: Jaypee Brothers Medical Publishers; 2010. p. 308.
3. Bertrand B, Eloy P, Cornelis JP, Gosseye S, Clotuche J, Gilliard C. Juvenile aggressive cement-ossifying fibroma: case report and review of the literature. *Laryngoscope* 1993;103:1385-1390.
4. Hamner JE 3rd, Lightbody PM, Ketcham AS, Swerdlow H. Cemento-ossifying fibroma of the maxilla. *Oral Surg Oral Med Oral Pathol* 1968;26:579-587.
5. Sarwar HG, Jindal MK, Ahmad SS. Cemento-ossifying fibroma—a rare case. *J Ind Soc Pedo Prev Dent* 2008;26:128-131.
6. Kos M, Luczak K, Godzinski J, et al. Treatment of monostotic fibrous dysplasia with pamidronate. *J Craniomaxillofac Surg* 2004;32:10-15.
7. Neville BW, Damm DD, Allen CM, Bouquot JE. Oral and maxillofacial pathology. Philadelphia: WB Saunders Co; 1995; 469-470.
8. Martín-Granizo R, Sánchez-Cuellar LA, Falahat F. Cemento-ossifying fibroma of the upper gingivae. *Otolaryngol Head Neck Surg* 2000;122:775.
9. Feller L, Buskin A, Raubenheimer EJ. Cemento-ossifying fibroma: case report and review of the literature. *J Int Acad Periodontol* 2004;6:131-135.
10. Sapp JP, Eversole LR, Wysocki GP: Contemporary oral and maxillofacial pathology. 2nd ed. St Louis, Missouri: Mosby; 2004. p. 116-117.
11. Barberi A, Cappabianca S, Colella G. Bilateral cemento-ossifying fibroma of the maxillary sinus. *Br J Radiol* 2003;76: 279-280.
12. Regezi JA, Sciubba JJ. Oral pathology-clinical pathologic correlations. 3rd ed. Philadelphia: WB Saunders Co; 1999. p. 357-360.
13. Neyaz Z, Gadodia A, Gamanagatti S, Mukhopadhyay S. Radiographical approach to jaw lesions. *Sing Med Pict Essay J* 2008;49(2):165.
14. Booth PW, Schendel SA, Hausamen JE: Maxillofacial surgery. 2nd ed. St. Louis, Missouri: Churchill Livingstone 2007; 506-509.
15. Brannon RB, Fowler CB. Benign fibro-osseous lesions: a review of current concepts. *Adv Anat Pathol* 2001;8:126-143.
16. Slootweg PJ, Muller H. Juvenile ossifying fibroma: report of four cases. *J Craniomaxillofac Surg* 1990;18:125-129.
17. Eversole LR, Leider AS, Nelson K. Ossifying fibroma: a clinicopathologic study of sixty-four cases. *Oral Surg Oral Med Oral Pathol* 1985;60(5):505-511.
18. Commins DJ, Tolley NS, Milford CA. Fibrous dysplasia and ossifying fibroma of the paranasal sinuses. *J Laryngol Otol* 1998;112(10):964-968.
19. Sakoda S, Shiba R, Irino S. Immediate reconstruction of the mandible in a patient with ossifying fibroma by replantation of the resected segment after freezing. *J Oral Maxillofac Surg* 1992;50:521-524.