

Cemento-ossifying fibroma with mandibular fracture. Case report in a young patient

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Abstract

The cemento-ossifying fibroma is classified as an osteogenic neoplasm of the jaws. It commonly presents as a progressively growing lesion that can attain an enormous size with resultant deformity if left untreated. A case of a large cemento-ossifying fibroma involving the left mandible is described in a 15 year old male patient. The clinical, radiographic and histological features as well as surgical findings are presented. The treatment of choice of this lesion is also emphasized. Two years after surgery, there was no evidence of recurrence and the trans-osseous wire used to immobilize the fracture was found to be completely buried in the jaw bone.

Key words: Cemento-ossifying fibroma, mandibular fracture, jaw neoplasm, case report.

(Received for publication April 1995. Accepted August 1995.)

Introduction

The cemento-ossifying fibromas or ossifying and/or cementifying fibromas have been described as demarcated or rarely, encapsulated neoplasms consisting of fibrous tissue containing varying amounts of mineralized material resembling bone and/or cementum.¹ These benign fibro-osseous lesions can arise from any part of the facial skeleton and skull with over 70 per cent of cases arising in the head and neck region.^{2,3} These cases involve mainly the mandible and maxilla but occasionally, they are reported in the orbitofrontal bone, nasopharynx, paranasal sinuses and skull base.²⁻⁴ It is not always easy to diagnose and manage the fibro-osseous lesions in the mandible or maxilla because their clinical, radiographic and histologic criteria often overlap causing confusion to clinicians, radiologists, pathologists and oral surgeons. This can result in differences of opinion and management of these lesions.

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Fibro-osseous lesions of the jaws were classified by Waldron⁵ initially into three main categories namely, fibrous dysplasia, fibro-osseous (cemental) lesions such as ossifying and cementifying fibroma, and fibro-osseous neoplasms such as juvenile active ossifying fibroma. In recent years, these 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 cemento-ossifying fibroma.⁶ In contrast, in the nomenclature by Kramer *et al.*¹ the cemento-ossifying fibroma is described as an osteogenic neoplasm and the fibrous dysplasia as a non-neoplastic bone lesion. Unlike fibrous dysplasia, the cemento-ossifying fibroma is well-circumscribed from its surrounding bone and this lesion will continue to grow bigger, slowly or actively, until it is removed surgically. Large lesions increasing in size to over 80 mm in their greatest diameter have been termed 'giant ossifying fibroma'.^{7,8} A small lesion of the lower jaw can be enucleated without difficulty, but a larger lesion involving the inferior border of the mandible⁹ or exhibiting a rapid growth¹⁰ is sometimes treated radically using bone grafts. The recurrence of these benign tumours following surgery is considered rare. However, Eversole and his co-workers¹¹ in a study of 64 cases of cemento-ossifying fibroma reported a recurrence rate of as high as 28 per cent following surgical curettage of these lesions. The cemento-ossifying fibromas are usually solitary, but bilateral¹² as well as multiple familial ossifying fibromas¹³ occurring in the jaws have been reported. In this paper, a case of a large cemento-ossifying fibroma occurring in a young patient and presenting with a mandibular fracture during surgery is described.

Case report

A 15 year old male Chinese, who complained of a swelling on the left side of his face, was referred by a private Consultant Radiologist to the Department of



Fig. 1.—Clinical appearance of patient at initial examination. Note, bowing of left mandible.

Oral and Maxillofacial Surgery, Faculty of Dentistry, Kuala Lumpur, for treatment of a large dentigerous cyst associated with the mandibular third molar. The swelling had been present for the past three months. It was asymptomatic but progressively increasing in size. No previous operation had been carried out at the site of the lesion and his past medical and family histories were non-contributory. The patient was generally healthy but an extra-oral examination revealed a slight facial asymmetry with a hard, non-tender bony swelling of the left mandible (Fig. 1). The overlying skin was intact with no anaesthesia noted on his lower left lip. The regional lymph nodes were not palpable. Intra-orally, the localized hard, bony swelling extended from the first premolar tooth to the third molar region with buccal and lingual expansion. The occlusion was satisfactory but the lower left first molar tooth was mobile. The overlying oral mucosa

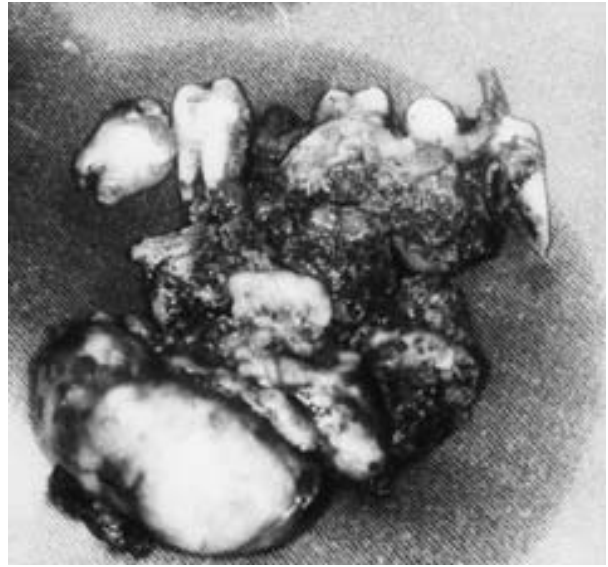


Fig. 3.—Cemento-ossifying fibroma has a smooth surface and is well demarcated from the bone.

was intact and normal in appearance. The teeth were all vital.

Radiographic examination revealed a multilocular and well-delineated radiolucent lesion measuring 65 mm×45 mm and presenting with faint patchy radiopacities (Fig. 2). There was evidence of root resorption in the first molar, displacement of involved teeth and erosion of the inferior cortex of the mandible. A pre-operative diagnosis of a fibro-osseous lesion such as a cemento-ossifying fibroma or fibrous dysplasia was made. Under local anaesthesia, an incisional biopsy was taken at the most expanded part of the buccal bone plate for histopathological examination. Laboratory investigations were also carried out to examine the alkaline phosphatase, serum calcium and inorganic phosphate levels of the patient.

The biopsy specimen taken indicated the tumour to be a cemento-ossifying fibroma. The laboratory tests showed elevated alkaline phosphatase, 244 IU/L

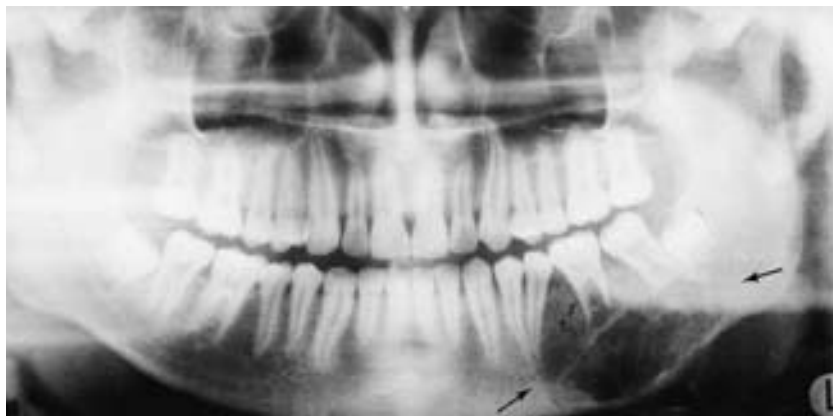


Fig. 2.—Well-defined radiolucent lesion with patchy mineralization (arrows).



Fig. 4.—Clinical appearance after two years with no recurrence.

(reference range, 34 to 135 IU/L); serum calcium, 3.0 mmol/L and phosphate, 0.6 mmol/L were within normal limits.

Surgical procedure

A conservative surgical approach was carried out under general anaesthesia to remove the large tumour. A flap was raised buccally and lingually to expose the bone from the distal of the left mandibular canine tooth to the retromolar region. The tumour mass was exposed by unroofing it occlusally. This was followed by sectioning the lesion into several segments and curetting it. At surgery, several sectionings were performed before the tumour together with the posterior teeth involved, could be enucleated completely (Fig. 3). Upon its removal, it was noted that there was a fracture on the body of the mandible as anticipated due to the very thin bone surrounding the tumour. Simple trans-

osseous wiring utilizing a 0.5 mm diameter, soft stainless-steel wire was carried out to immobilize the fracture. At the tumour site, the remaining sound bone was smoothed and curetted with a large acrylic bur. The cavity was then packed with ribbon gauze soaked in Whitehead's varnish to fill the osseous defect.

The patient was reviewed regularly during the postoperative period and the pack was changed every two weeks until bone had filled up the defect. After eight months, the wire used was found to be completely embedded in the jaw bone. A two-year follow-up period showed the wire to be present near the mid-point of the bone and the bowing contour of the mandible had improved slightly but did not disappear (Fig. 4). There was no evidence of recurrence and the patient had no problem using a lower denture (Fig. 5).

Histopathology

Macroscopic findings

The specimen consisted of a large hard solid mass (Fig. 6) measuring 50×50×30 mm, all the left mandibular posterior teeth, and six pieces of hard tissue measuring 30×30×20 mm. These tissues were decalcified and routinely processed for histopathology.

Microscopic findings

Histological examination of the lesional tissue showed a well-circumscribed and partly encapsulated fibro-osseous lesion consisting of an abundant cellular fibrous tissue with scattered trabeculae of lamellar bone, metaplastic/woven bone and cementoid masses (Fig. 6, 7). The stromal cells were spindle-shaped and had bland-looking nuclei. The erupted teeth and another unerupted partially formed molar exhibited normal dentine, enamel and pulp. In the latter tooth, the dental follicle contained focal areas of epithelial cell rests.



Fig. 5.—Radiograph shows wire completely embedded in bone.

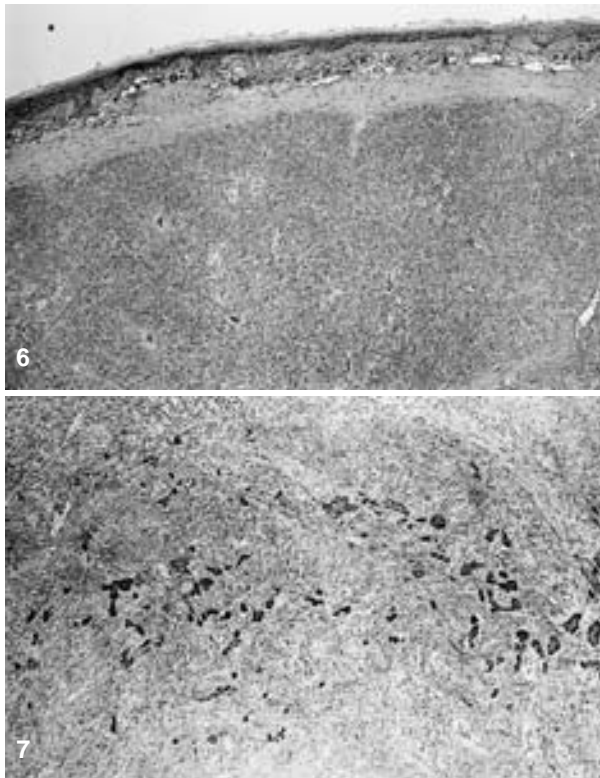


Fig. 6.—Peripheral circumscription and partial encapsulation. H & E $\times 40$.

Fig. 7.—Lesional tissue consists of cellular fibrous stroma containing scattered trabeculae of metaplastic bone and cementoid material. H & E $\times 100$.

Correlating clinical, radiological and histological findings, the case was reported as a benign fibro-osseous lesion consistent with cemento-ossifying fibroma.

Discussion

The term cemento-ossifying fibromas has been used to refer to all the fibro-osseous lesions previously classified as ossifying and/or cementifying fibromas as they fall within the spectrum of the same disease entity. Waldron and Giansanti¹⁴ believed that the presence and ratio of the osteoid and cementum reflected this spectrum of the disease. Cementum-like substances of these lesions have also been found in other facial bones such as the maxillary antrum, sphenoid, orbitofrontal and temporal bones¹⁵ away from the tooth-bearing mandible or maxilla. Eversole *et al.*¹¹ reported that the production of these cementum-like structures may be associated with membranous bone and may not only be related to cementogenesis.

Clinically, the cemento-ossifying fibroma presents as a painless, slowly increasing expansion of the jaw. It occurs mainly in the second to the fourth decades of life and is more common in women than men.^{2,6,16} The majority of the lesions arise from the premolar-molar region and appear to be more aggressive in

younger patients.^{2,17,18} The words 'juvenile', 'active' and 'aggressive' are used commonly to refer to the age of the patient, the growth rate of the lesion and its tendency to recur, respectively. In this case the tumour arose from the posterior part of the mandible in a young patient. The progressive increase in size of this tumour over a period of three months with resultant facial asymmetry correlated well with the clinical characteristics of this entity.

Radiographically, it commonly presents as a well-circumscribed radiolucent area with a speckled appearance and may be unilocular or multilocular in pattern.¹¹ When the lesion is large, there may be root resorption, displacement of teeth in the tooth-bearing region, and in the mandible, erosion of the inferior cortex.^{5,9} Most of these features were seen in the radiographs taken of this lesion with the exception that the shape of the cemento-ossifying fibroma reported here was multilobular when excised surgically rather than multilocular as previously described.

Histologically, different hard-tissue configurations can be observed but this makes no difference to the clinical behaviour of the tumour. However, recognition of these structures is important in establishing its diagnosis. They consist of an admixture of cementum-like material and metaplastic bone.¹ Both components were identified in the current lesion. At the microscopic level, the principal differential diagnosis considered for this case included fibrous dysplasia, juvenile (aggressive) ossifying fibroma and benign osteoblastoma. Fibrous dysplasia was ruled out based upon the histological demonstration of circumscription and partial encapsulation of this lesion because these are not features of fibrous dysplasia as the lesional bone in the latter tend to fuse directly with the surrounding jaw bone.¹ Juvenile (aggressive) ossifying fibroma was considered and excluded because the histological presentation of this entity was significantly different from what was observed in the current case. Unlike the cemento-ossifying fibroma, the juvenile (aggressive) ossifying fibroma characteristically consists of a cell-rich fibrous tissue containing cellular osteoid, woven bone, small foci of giant cells and sometimes abundant osteoclasts related to the woven bone.¹ These features were not observed in the multiple sections of the enucleated specimen of this case. The benign osteoblastoma may also present as a well-circumscribed lesion histologically. However, unlike the cemento-ossifying fibroma which is a relatively avascular fibro-osseous lesion, the benign osteoblastoma is a vascular, osteoid-forming neoplasm.¹⁹ Microscopic characteristics of this entity include a highly vascular and cellular tissue containing numerous trabeculae of osteoid with osteoblast rimming and immature bone with varying degrees of calcification. A few multinucleated giant cells may

be present. These features were not encountered in the current lesion.

Treatment of cemento-ossifying fibroma generally has been by conservative enucleation/curettage or radical surgery. Sakoda *et al.*²⁰ described the procedure of a segmental resection of an extensive ossifying fibroma with the replacement of the excised segment after cryotherapy. Sloomweg and Muller,²¹ however, reported that there was no difference in outcome between patients treated in a more limited way and those treated by major surgery. Conservative surgery is therefore recommended even if the tumour is large with bowing and erosion of the inferior border of the mandible as shown. Radical treatment of the tumour such as an *en bloc* resection should only be considered if there are recurrences due to its aggressive nature. There are several advantages for treating large cemento-ossifying fibromas conservatively. For example, there is minimal morbidity after surgery, good bone formation and consolidation, no loss of sensation as well as no bone graft required from a second surgical site. In long term follow-up cases where bowing or contours formed by these large lesions do not disappear completely with time, surgical intervention such as aesthetic recontouring of the bone may then be taken into consideration or recommended. In the current case, in view of the patient's young age, acceptable present aesthetic facial features and improved contour of the mandible, a further cosmetic operation was not considered. Good results in regard to cosmetic and functional deformity can be achieved using this approach for a large cemento-ossifying fibroma.

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