Cementifying fibroma of the maxillary sinus: a case report

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ABSTRACT

A case of cementifying fibroma of the maxillary sinus that occurred in a 25-year old Saudi male is reported. Diagnosis depends upon clinical, radiographic and histological evaluation of an incision biopsy. The uncommon location of the reported tumor in the maxillary sinus probably accounts for its large size, aggressive behavior and osseous destruction. Prognosis was excellent after complete enucleation. In reporting the case, differential diagnosis of fibro-osseous lesions in the maxilla has been reviewed.

Keywords: Cementifying fibroma, clinical appearance, histology, radiographical appearance, differential diagnosis, treatment.


Cementifying fibroma is a histological term used to describe a benign tumor affecting the jaws, showing fibroblastic tissue containing rounded or lobulated masses of calcification which have no relation to the apex of the tooth. It is one of the fibro-osseous lesions, first recognized by Brophy and described in detail by Staffe. Based on the work of Pindborg and Kramer, the World Health Organization classified cementomas into four types: cementifying fibroma, gigantiform cementoma, cementoblastoma and periapical cemental dysplasia. Cahn and Tiecke considered that cementifying fibroma is probably a variant of osseous fibroma, the only difference between the two lesions being that some pathologists interpret the hard tissue in the former lesion as cementum and in the latter as bone. If the lesion contains both cementum and bone it is called cemento-ossifying fibroma. Although cementifying fibroma affects the jaws, it uncommonly occurs in the maxillary sinus, and very rarely in ectopic sites.

Case Report. A 27 year old Saudi male patient was referred to the Oral and Maxillofacial Unit at the Riyadh Armed Forces Hospital from the Primary Care section, complaining of swelling affecting the left side of the face, and blockage of the left nostril which had worsened gradually over the previous two months. Past dental history revealed that the upper left first molar had been extracted due to caries five years previously without any difficulties. The associated teeth were moderately loose.

On examination there was a localized swelling of the left cheek as shown (Figure 1 - permission was given from patient to be photographed) and complete blockage of the left nostril with nasal congestion. No lymph nodes were palpable. Intra-oral examination showed a swelling affecting the maxillary left buccal sulcus and extending from the canine to the third molar region. A palatal swelling related to the same region was also noticed extending to mid-palatal area (Figure 2). Orthopantomograph and sinus views showed a radiopaque shadow filling the left sinus. In the computerized tomography (CT) (Figure 3), there was a well-defined rounded lesion filling the left maxillary sinus, extending medially to the nasal septum and anterolaterally and inferiorly to cause resorption of the floor of the sinus. The lesion was
close to the posterior wall of the sinus.

Incision biopsy of the left sinus was performed through an intra-oral approach under local anaesthesia. Histopathological examination demonstrated dense fibroblastic tissue with regularly scattered foci of cementum. No evidence of malignancy was reported, with the specimen being reported as cementifying fibroma.

Treatment consisted of surgical excision under general anaesthesia through the CaldwellLuc approach. An incision was made medial to the upper left canine, extending back to the upper left third molar, and the flap was reflected superiorly. There was a defect in the antero-lateral wall of the maxillary sinus with the lesion bulging through the buccal palate. The upper left second premolar and second and third molars were extracted. The lesion was firm, compressible and rubbery which made its enucleation possible without rupture. The lesion measured 4.5 x 5 x 6 cm. The defect was packed with one inch ribbon gauze with bismuth iodine paste. The end of the pack was accessible by virtue of its placement through the left nostril after antrostomy. The wound was closed using 3/0 vicryl sutures, and post-operative oral augmentin 375 mg t.d.s., with codine phosphate 60 mg intramuscularly as requested, were prescribed. Post-operatively, the patient had immediate relief from the nasal

Figure 1 - The photograph shows a swelling of the left cheek causing elevation of the left eye. Note the difference in the sclera between the two eyes. Permission was given from patient to be photographed.

Figure 3 - The sagittal section of the CT shows the space occupied by the lesion in the left maxillary sinus. The lesion extends medially to block the left nasal cavity.

Figure 2 - The photograph shows the extension of the tumor to the buccal sulcus and palate. The suture represents the site of biopsy.

Figure 4 - Histopathology section showing plenty of fibrous tissue, cementum and few lamellar bone (Hematoxylin and erythrosin stain magnification x 100).
obstruction. The antral pack was removed two days post-operatively after the patient had received pethidine 75 mg intramuscularly. One year follow-up post-operatively revealed no complaints and no signs or symptoms of recurrence.

**Discussion.** Cementifying fibroma is a benign tumor and considered as one of the fibro-osseous lesions of the jaws,\(^1\) in which normal bone is replaced by cellular fibroblastic tissue containing rounded or lobulated masses of calcified material in the form of cementum.\(^3\) If the calcified material is in the form of bone, the lesion is called ossifying fibroma, while if it is a mixture of bone and cementum it is called cemento-ossifying fibroma. However, it is doubtful whether any significant difference in biologic behavior between these lesions and cementifying fibroma exists.\(^3\) Although the origin of the tumor is not clear, there is universal agreement,\(^4-5\) that the tumor is a product of aberrant periodontal membrane growth and development. The multipotential cells in the periodontal membrane are capable of forming fibrous tissue, cementum and lamellar bone.

The triggering mechanism which initiates the formation of cementum outside the periodontal membrane is unclear, but may include infection, trauma or dental extraction. Under these conditions the progenitor cells are capable of producing cementum, bone and fibrous tissue, and the resultant tumor contains cementum or lamellar bone or fibrous tissue or any mixture of these components in varying amounts. Dehner\(^6\) felt that the predominant pattern determines the final classification despite a varying degree of morphologic overlap. There are three developmental stages in cementifying fibroma: the first, the osteolytic phase, is characterized by cellular tissue formation with no calcified material deposits. This represents an immature cementoma and appears radiolucent on X-ray. In the second stage, there is cementum deposited in the fibrous mass which becomes relatively calcified and appears radiographically as a mixture of radiolucence with some radiopaque patches. In the final stage, the entire mass becomes calcified and is encapsulated by a remnant of connective tissue; this is called the inactive stage because usually there is no increase in size while on X-ray it appears as a radiopaque mass.

Clinically, cementifying fibroma is more common in the mandible, especially in the premolar and molar region, than in the maxilla. However, in the reported tumor inspite of the fact that the tumor filled the entire maxillary sinus, its area of origin may be the alveolar ridge of the upper first molar which had been extracted five years earlier since it was suggested in the literature that dental extraction may leave part of the periodontal membrane attached to the wall of the alveolus. Cementifying fibroma affects females more than males and usually occurs between 30 and 40 years of age. It is a slowly-growing tumor but in some cases the tumor grows rapidly,\(^10,14\) and it is asymptomatic until it reaches considerable size causing pressure on surrounding structures, which in turn reflects the nature of the signs and symptoms. Lesions in the maxillary sinus can cause nasal blockage, proptosis, epiphora, looseness of the adjacent teeth, obliteration of buccal sulcus and facial swelling.\(^7,10,15\) In the present case the tumor seems to have expanded in all directions to cause facial and palatal swelling, nasal blockage and looseness of the associated teeth.

Radiological appearance reflects the stage of development, and consequently it may be anything from radiolucent to radiopaque.\(^1,2,16\) Being a benign tumor and relatively slow growing compared to the malignant tumor, there is remodeling of bone by the periosseum at a rate similar to that of the bone erosion by the mass; there is also a rim sign which is characteristic of the benign tumor.\(^17\) In the case reported here, the sagittal section of CT (Figure 2) was very useful to show the extent of the lesion and its relation to the surrounding structures.

Histologically it is not easy to distinguish between bone and cementum and from the clinical and treatment stand points there is no difference between cementifying or ossifying fibroma.\(^18\) The surface was smooth and covered by a capsule, and the cut surface was yellowish white in color. Microscopically it showed mainly dense fibroblastic tissue with few regularly scattered foci of cementum, most of which was deeply basophilic and resembling cementum; there was also a small amount of bone (Figure 4). Differential diagnosis included fibrous dysplasia, myxoma, calcifying cyst, adenomatoid odontogenic tumor, calcifying epithelial odontogenic tumor, Paget's disease and desmoplastic fibroma.

The aim of treatment is not only to remove the tumor but also to minimize the risk of its recurrence, preserve vital structures and to maintain good function and aesthetics. Yet, there is a universal agreement that conservative treatment of the lesion by enucleation is adequate to achieve these aims.\(^1,5,6,10,20\) However, some authors feel that, in cases of rapid growth, more aggressive surgery may be necessary.\(^12\) Radiation is contraindicated due to its radio-resistance and high risk of malignant transformation.\(^7\) Even in cases of extension of the tumor to vital structures like the orbit, sacrifice of the orbit is not recommended.\(^10,27\) Prognosis is good and, although recurrence is very rare,\(^1,5\) it is difficult to be estimated from the available literature, since most reports are of only one or two cases. However, Hamner et al\(^28\) reviewed 67 cases and showed only three cases of recurrence after initial surgical removal, while Taylor\(^29\) showed one case of recurrence after 24 years.
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References


