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Central Cementoossifying Fibroma of the Maxillary Sinus: A Review of Six Cases

A. John Kuta, C. MacDonald Worley, and George E. Kaugars

Summary: We present the radiographic findings of six patients with central cementoossifying fibromas of the maxilla. CT typically demonstrated large, spherical tumors in the maxillary alveolar ridge, filling and expanding the maxillary sinus and extending to involve the ipsilateral hard palate. The central tumors ranged from having soft-tissue density with scattered foci of high density to being heavily calcified.

Index terms: Paranasal sinuses, fibroma; Maxilla

Central cementoossifying fibromas (also known as central ossifying fibromas or central cementifying fibromas) are uncommon benign fibroosseous lesions of the jaw thought to originate from the periodontal ligament. Most are small and incidentally diagnosed with routine dental radiographs. With larger lesions, patients may complain of an abnormal bite or an enlarging mass. We present our experience with six cases of central cementoossifying fibroma of the maxilla.

Materials and Methods

The clinical, radiographic, and pathologic data of six patients with central cementoossifying fibromas of the maxilla evaluated in the past 5 years at our institution were reviewed. All patients were evaluated with computed tomography (CT) at our institution or a referring hospital. One patient also was studied with magnetic resonance (MR) (Siemens Magnetom 1.5-T, Iselin, NJ). Five of these six tumors were totally excised; one was biopsied and is awaiting surgery. The pathologic specimens were interpreted by one oral pathologist (G.E.K.).

Results

Each of six patients presented with either an abnormal bite or an enlarging facial mass. Plain radiographs demonstrated an opacified maxillary sinus with elements of increased density. CT typically demonstrated a large, spherical tumor originating in the maxillary alveolar ridge. The mass filled and expanded the maxillary sinus. The tumor commonly extended to involve the ipsilateral hard palate. The central tumor ranged from soft tissue density with scattered foci of high density to heavily calcified. A thin shell of bone was present around each tumor without cortical bone erosion. In the one patient imaged with MR, the central cementoossifying fibroma had homogeneous signal isointense to muscle on T1-weighted images and very hypointense on T2-weighted images. The clinical and radiographic features of each patient are presented in the Table.

Case 1

Patient 1 is a 31-year-old man who presented to his dentist with a "bad bite" and right nasal obstruction. Physical examination revealed a 4 \times 4-cm firm, nontender, asymmetric expansion of the right hard palate and maxilla. There was no facial anesthesia. Extraocular eye muscle movements were normal. A plain radiograph of the maxillary sinus demonstrated opacification of the right maxillary sinus. CT demonstrated a $4 \times 5 \times 5$ -cm, well-defined, mixed-density mass filling the right maxillary sinus (Fig 1). The mass extended from the nasal septum and hard palate to the lateral maxillary sinus wall, from the anterior to the posterior wall of the maxillary sinus, and from the maxillary alveolar ridge to the orbital floor. The posterior wall of the sinus and orbital floor were displaced. A biopsy of the lesion was diagnosed as a central cementoossifying fibroma. Complete surgical

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Radiologic features of maxillary central cementoossifying fibromas

Patient	Age, y/Sex	Presenting Symptom	Size of Central Cementoossifying Fibroma, cm	Dense Foci in Central Cemento- ossifying Fibroma	Nasal Septum Involvement	Orbit Floor Involvement	Palate Extension	Recurrence/ Duration of Follow-up
1	31/M	"Bad teeth"	4×5×5	+	+	+	+	None/3 years
2	19/F	Expansion of maxilla noted by dentist	2×3×3	_	_	_	+	None/4 years
3	19/F	Facial swelling	$3\times4\times5$	+	-	+	+	None/5 years
4	27/M	Facial swelling	$4\times4\times4$	+	_	_	+	Not yet excised
5	47/F	Ill-fitting dentures	$3\times3\times4$	+	_	_	_	None/1 year
6	37/M	Facial swelling	3×4×5	+	_	_	+	None/2 years

Note.— + indicates present; -, not present.

excision was performed through a transoral approach. There has been no recurrence after 2 years.

Case 2

Patient 4 is a 27-year-old man who presented with right facial swelling. There was expansion of the hard palate from the mass (Fig 2A). A bitewing radiograph showed divergence of the roots of the teeth (Fig 2B). A core biopsy diagnosed a central cementoossifying fibroma. CT performed 2 weeks after the biopsy showed a large mass expanding into the maxillary sinus with multiple small foci of calcification (Fig 2C and D). MR examination demonstrated the mass to be isointense to muscle on T1-weighted images and very hypointense on T2-weighted images (Fig 2E and F).

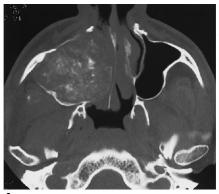
We have imaged another patient with a clinical diagnosis of central cementoossifying fibroma who demonstrated CT and MR imaging characteristics identical to those listed above. Because we do not have histologic proof of the

diagnosis, we have not included her in our results.

All six tumors demonstrated similar histopathologic characteristics. Osteoid, bone, and cementum were contained within a hypercellular fibrous connective tissue stroma. The fibrous connective tissue had occasional mitotic activity, but abnormal cells were not evident (Fig 3).

Discussion

Central cementoossifying fibromas are a distinct form of benign fibroosseous lesions of the mandible and maxilla. They are thought to arise from the periodontal ligament and are composed of varying amounts of cementum, bone, and fibrous tissue. Cementum is the mineralized connective tissue that covers the root of the tooth. The hybrid name *central cementoossifying fibroma* is used because there is a spectrum of fibroosseous lesions that arise from the periodontal ligament, ranging from those with only deposition of cementum to those with only deposition of bone (1). Central cementoossifying fibromas occur more frequently in women than



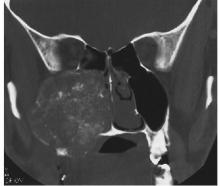


Fig 1. Patient 1. A, Axial and B, coronal CT of the maxilla demonstrate a large central cementoossifying fibroma filling the maxillary sinus with diffuse foci of calcification and expansion of the sinus walls. The mass extends to the orbital floor and nasal septum. The hard palate is involved.

A B

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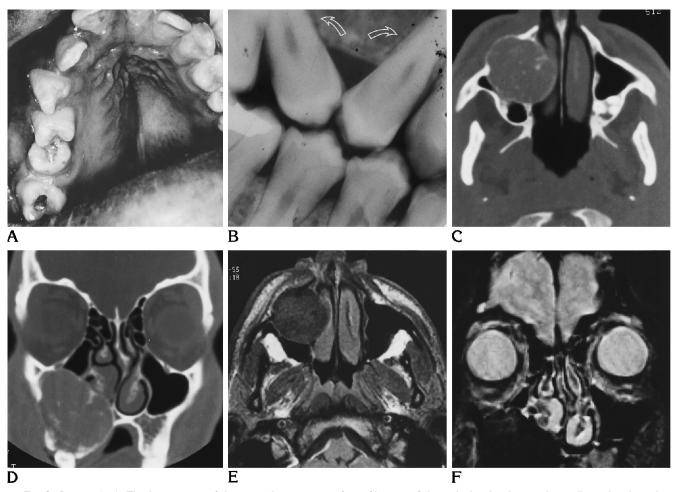


Fig 2. Patient 4. A, The large mass of the central cementoossifying fibroma of the right hard palate and maxillary alveolar ridge expands the hard palate beneath the mucosa.

B, Bitewing radiograph shows divergence of the first and second right maxillary premolars by the mixed-density mass of the central cementoossifying fibroma (*arrows*).

C, Axial and D, coronal CT scans show the central cementoossifying fibroma to have scattered foci of dense calcification. The margin is well defined and sclerotic. It involves the maxillary sinus mass and the hard palate.

E, Axial T1-weighted MR image shows the homogeneous signal of the mass, isointense to the pterygoid muscles.

F, Coronal first echo of T2-weighted image demonstrates the extreme hypointense signal of the central cementoossifying fibroma in the right maxillary sinus.

in men. They arise in the mandible in 62% to 89% of patients, 77% occurring in the premolar region. Most are diagnosed between 20 and 40 years of age (2, 3). When this tumor arises in children, it has been named the *juvenile aggressive cementoossifying fibroma*, which presents at an earlier age and is more aggressive clinically and more vascular at pathologic exam (4). Central cementoossifying fibromas are asymptomatic until they cause expansion. Thus, they are generally not diagnosed until the tumor has had time to produce calcifications. One remarkable finding is the large size of the maxillary tumors in our series at the time of diagnosis, probably attributable to the large amount of

available space in the maxillary sinus into which they could expand.

Although central cementoossifying fibromas of the mandible are common, central cementoossifying fibromas of the maxillary sinus are unusual tumors; 25 have been reported in the literature (4). Central cementoossifying fibromas are typically well-defined, solitary radiolucencies with scattered radiopaque foci. They vary in radiopacity depending on the amount of cementum and bone that have been deposited (Fig 4). They maintain a spherical shape, expand the surrounding cortical bone without cortical perforation, and may cause tooth divergence. Large tumors may involve the nasal

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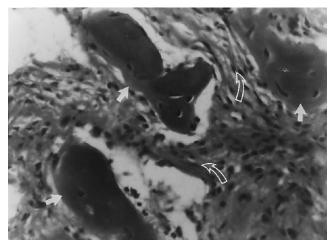


Fig 3. Histologic examination reveals irregularly shaped fragments of osteoid and cementum ($closed\ arrows$) within a hypercellular fibrous connective tissue stroma ($open\ arrows$). (Hematoxylin and eosin stain, magnification $\times 80$)

septum, orbital floor, and infraorbital foramen. The tumor extent guides surgical therapy. Maxillary central cementoossifying fibromas are large at the time of presentation, indicating the capacity of the tumor to expand freely within the maxillary sinus. The degree of involvement of the other structures of the face are listed in the Table.

The central cementoossifying fibroma imaged with MR was isointense to muscle on T1 and had a diffuse homogeneous low signal on T2. This low signal likely represents the low free-water content of the calcific and fibrous tumor.

Pathologic examination of the central cementoossifying fibroma shows a proliferation of ir-

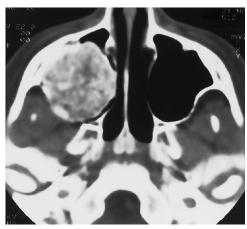


Fig 4. The range of mineralization within central cementoossifying fibromas is demonstrated by the heavy mineralization of patient 3 with this axial CT when compared with the sparse mineralization seen in Figure 1.

regularly shaped calcifications within a hypercellular fibrous connective tissue stroma. The calcifications are extremely variable in appearance and represent various stages of bone and cementum deposition. Histologic differentiation between osteoid and cementum is difficult. In some cases, most of the calcified fragments are immature cementum, with basophilic coloration on hematoxylin and eosin-stained sections. These tumors have been named central cementifuing fibroma. In other cases, the calcified fragments are osteoid, with typical eosinophilic coloration on hematoxylin and eosin-stained sections. These tumors have been named central ossifying fibromas. However, central ossifying fibromas also can be basophilic, causing difficulties with differentiating from central cementifying fibromas. Most pathologists feel that central cementifying fibromas and central ossifying fibromas arise from the same progenitor cell but produce variable amounts of bone and cementum within any one lesion. The hybrid term central cementoossifying fibroma has evolved to indicate the likely presence of both types of tissue within the same lesion because of the difficulty in being able to distinguish reliably immature bone from immature cementum and because of the presence of both of these substances in many of the lesions. Thus, central cementoossifying fibroma is the most accurate histologic term, but it can be interchanged with either central ossifying fibroma or central cementifying fibroma. There is no apparent clinical or radiologic difference between the central cementifying fibroma or central ossifying fibroma, so the hybrid central cementoossifying fibroma works well for radiology, too.

Maxillary central cemento-ossifying fibromas tend to display a greater degree of immaturity than that seen in mandibular lesions, but there is no reliable pattern useful to distinguish between maxillary and mandibular lesions. There is a correlation between the amount of calcification seen in the surgical specimen and that seen on the CT. The pathologic differences between central cementoossifying fibroma and fibrous dysplasia are few and the diagnosis must be made in light of the radiographic findings.

The differential diagnosis includes other lesions that contain radiopacities within a well-defined radiolucent mass: chondrosarcoma or osteosarcoma, fibrous dysplasia, odontogenic cysts, squamous cell carcinomas, calcifying

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odontogenic cysts (Gorlin cysts), and calcifying epithelial odontogenic tumors (Pindborg tumors). The well-defined border of the central cementoossifying fibroma helps differentiate it from the aggressive sarcomas and carcinomas. Fibrous dysplasia has a characteristic "ground glass" appearance not seen in the central cementoossifying fibroma. The radiologic differentiation of central cementoossifying fibroma from Gorlin cysts and Pindborg tumors is difficult; the final diagnosis is based on histologic appearance. Pindborg tumors have a high association with impacted teeth.

The recommended treatment of the central cementoossifying fibroma is excision. The entire tumor should be removed including involved regions of the orbital floor and maxillary sinus walls. Central cementoossifying fibromas usually "shell out" easily at surgery, but maxillary central cementoossifying fibromas are more difficult to remove completely than mandibular central cementoossifying fibromas. This may be attributable to the difference in bone character between the mandible and maxilla and to the available space for expansion in the maxillary sinus. Recurrence has been reported in as many as 28% of patients with mandibular central cementoossifying fibromas. The recurrence rate of maxillary central cementoossifying fibromas is unknown, but it is likely to be higher because of the greater difficulty of their surgical removal and larger size at the time of presentation.

Conclusions

The central cementoossifying fibroma of the maxilla is an uncommon benign tumor. Cos-

metic and dental occlusal problems are often the first manifestations of these lesions. CT reveals a well-defined, mixed-density tumor with diffuse scattered calcification. It protrudes into the sinus, maintaining a thin bone shell that is a remnant of the alveolar process. The lesion can involve all the sinus walls, the hard palate, and the nasal septum. MR shows a mass with diffuse low signal consistent with the calcification. In the maxilla, the clinical and radiologic differential diagnosis includes fibrous dysplasia, osteosarcoma or chondrosarcoma, squamous cell carcinoma of the maxillary sinus, calcifying epithelial odontogenic tumors (Pindborg tumors), and calcifying odontogenic cysts (Gorlin cysts).

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