

Case Report

Cemento-Ossifying Fibroma Presenting as a Mass of the Parapharyngeal and Masticator Space

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Summary: We report a case of cemento-ossifying fibroma that presented as a large extraosseous mass in the masticator and parapharyngeal space. CT scanning and MR imaging showed a large extraosseous mass with central conglomerated, well-matured ossified nodules and fatty marrow. The central matured ossified nodules were of low density on CT scans and high signal intensity on T1- and T2-weighted MR images. Multiplanar reformatted CT scans revealed the origin of the mass to be at the extraction site of the right lower second molar tooth.

Cemento-ossifying fibroma (COF) is a well-demarcated and occasionally encapsulated neoplasm that contains fibrous tissue and varying amounts of calcified tissue resembling bone, cementum, or both. Ossification of this material is rare, and woven bone is predominant. This neoplasm occurs in patients of a wide age range, but the greatest numbers of cases are encountered during the third and fourth decades of life. There is definite female predilection, with female-to-male ratios as high as 5:1. The mandibular premolar-molar area is the most common site. COF of the head and neck is described radiographically as a well-circumscribed, expansile bony lesion with calcified matrices in the mandible and maxilla. Several rare cases involving the nasal bones, orbit, ethmoid sinus, sphenoid sinus, maxillary sinus, occiput, temporal bone, and nasopharynx have also been reported (1–6). However, to the best of our knowledge, there is no report in the English literature of ossifying fibroma that presented as an extraskelatal mass in the masticator and parapharyngeal space. We describe imaging and pathologic findings and a possible pathogenesis of extraskelatal COF.

Case Report

A 59-year-old woman was referred to our hospital because of right retromolar swelling and a 5-month history of pain.

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Her medical history was otherwise unremarkable. A clinical examination revealed a hard expansile mass that was palpable along the medial border of the right mandibular ramus. The right lower second molar tooth was absent. CT scans (Fig 1A–B) showed a large, lobulated, bone-density mass adjacent to the right mandibular ramus in the right masticator and parapharyngeal space, displacing the medial and lateral pterygoid muscles. Centrally heterogeneous low densities were seen. The medial cortex of the mandible was eroded at the site of tooth extraction. A multiplanar reformatted image showed the origin of the mass at the site of an extracted molar tooth (Fig 1C). T1- and T2-weighted MR images (Fig 1D–E) showed a heterogeneous low signal intensity. A central high signal intensity area on T1- and T2-weighted MR images corresponded to low attenuation on CT scans, suggesting mature bone with fatty marrow. After infusion of the contrast material, subtle enhancement was seen in the portion of the mass. The margin of the mass was indistinct on the MR images because signal intensity was similar to that of the adjacent muscles on T1- and T2-weighted images. A biopsy was performed with the patient under local anesthesia, and the results of a microscopic examination were consistent with COF (Fig 1F). Using a lip-splitting approach, a continuity resection with hemimandibulectomy through the first molar was performed. During the dissection, the lesion was easily separable from the adjacent lingual mucosa and displaced pterygoid muscles but was firmly attached to the bone. Reconstruction surgery was performed using the acrylic resin mold. Because of wound infection, the infected resin mold was removed and a second reconstruction was performed with the plate. No clinical sign of recurrence was observed for 10 months. A follow-up CT study to be conducted 1 year after tumor resection was planned.

Discussion

COF is a well-demarcated benign fibrous neoplasm that contains varying amounts of calcified tissue resembling bone, cementum, or both. In the past, many investigators separately classified cementifying fibromas from ossifying fibromas. When curvilinear trabeculae or spheroidal calcifications were encountered, the lesion was often referred to as cementifying fibroma. When bone was predominated, ossifying fibroma was assigned. Today, however, the term “cemento-ossifying fibroma” is widely used because both osseous and cemental tissues are seen commonly in a single lesion (7).

COF is characterized by a well-defined expansile bony mass without destruction and rarely has an extraosseous soft-tissue component. Our case had a small bony component and a large extraosseous component. The small bony component was located at the root of the extracted right lower second molar tooth and extended superiorly and medially with cortical destruction. The larger extraosseous

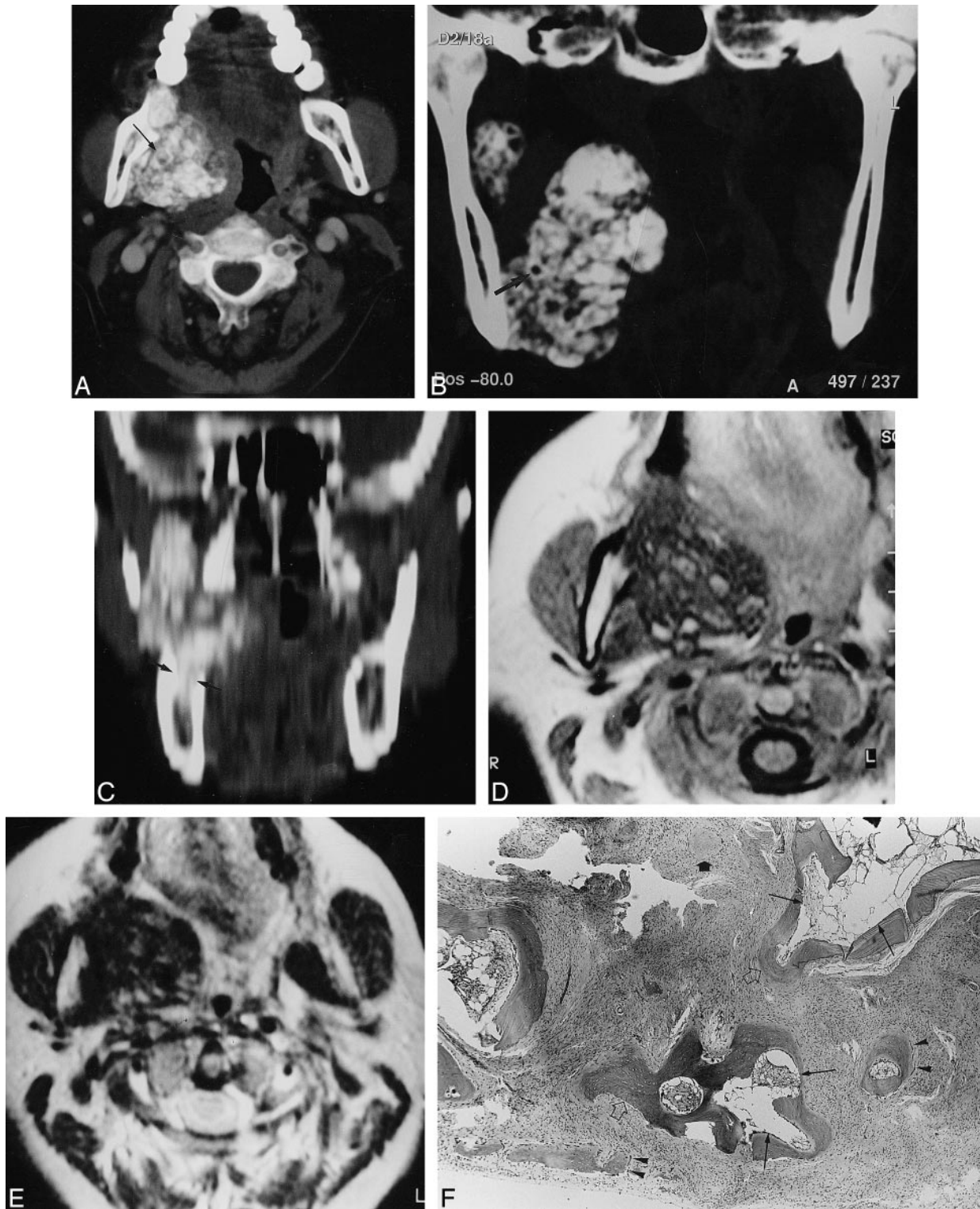


FIG 1. A 59-year-old woman presented with right retromolar swelling and a 5-month history of pain.

A, Axial CT scan shows a large lobulated bone density mass in the right masticator and parapharyngeal space. This mass is located in the intermuscular spaces between the medial and lateral pterygoid muscles. Central low-density areas (*arrow*) are seen.

B, Coronal CT scan shows a large lobulated bone density mass in the right masticator and parapharyngeal space. This mass is located in the intermuscular spaces between the medial and lateral pterygoid muscles. Central low-density areas (*arrow*) are seen.

C, Multiplanar reformatted coronal CT scan shows the origin of the mass at the extracted molar tooth site (*arrows*).

D, Axial T1-weighted (750/15 [TR/TE]) image shows a low-signal-intensity mass in the right masticator and parapharyngeal space. Multiple nodules within the mass show a low-signal-intensity rim and a central high signal intensity.

E, This central high signal intensity is hyperintense on T2-weighted images (2200/90) and corresponds to central low attenuation on the CT scan, suggesting mature bones with central fatty marrow.

F, Microscopic study shows that the tumor is composed of fibrous tissue containing an admixture of immature woven bone (*thick short arrow*) and mature lamellar bone (*open arrows*). Central fatty marrow (*long arrows*) is seen within the mature lamellar bone ($\times 40$). Osteoblastic rimming of trabeculae is also shown (*arrowheads*).

component was located in the intermuscular space between the medial and lateral pterygoid muscles.

The origin of COF is thought to be the periodontal membrane, a layer of fibrous connective tissue surrounding all tooth roots. The connective tissue of the periodontal membrane harbors the potential for elaboration of both bone and cementum. Bernier and Thompson (8) speculated that infection with resulting inflammation and fibrosis of the periapical area might stimulate the periodontal membrane. After trauma, such as tooth extraction, the remaining periodontal tissue that is attached to the wall of the alveolus may serve as the origin of COF. In our case, the tumor might have developed at the remaining periodontal membrane after tooth extraction. Upward growth and large extrasosseous mass formation might be related to lesser resistance after extraction of the second molar tooth. Several rare cases outside the mandible and maxilla have been reported. The current theories regarding their origin include traumatic and developmental causes. Cakir and Karadayi (1) suggested that nasopharyngeal COF originated from embryologic nests. Brademann et al (3) explained that ectopic periodontal membrane differentiated from primitive mesenchymal cells in the petrous bone may serve as a cause of development of COF in this area and that trauma such as severe whiplash may be a factor in the induction of proliferation of COF. The ethmoidal location of COF may also be explained by incomplete migration of mesenchyme and its differentiation into periodontal membrane.

The mass was well demarcated from the surrounding soft tissue and did not invade the adjacent muscles, although it extended along the intermuscular space. During the operation, it was separated easily from the displaced pterygoid muscles but was firmly attached to the bone. The nasopharynx and oropharynx were narrowed by the extrasosseous component of the mass.

On the CT scans, a major portion of the mass consisted of numerous ring-shaped and curvilinear ossified nodules. A central low-attenuation area on the CT scans corresponded to high signal intensity on T1- and T2-weighted MR images. These nodules were considered to be mature bone with central fatty marrow. Immature woven bone and fibrous connective tissue, as well as mature lamellar bone with the central fatty marrow, were evident after pathologic analysis. As the lesion matures, ossification increases and coalesces, accounting for the lesion's progressive increase in radiodensity. Ossification consists of immature woven bone that matures into lamellar bone.

This mass showed a different pathologic finding from that of common COF. Ossification of imma-

ture bone, which is rarely seen in cases of common COF, was a prominent feature of the lesion in our case. We hypothesize that because the mass was detected in a more mature person, the immature woven bone, typical of common COF, had progressed to form mature lamellar bone with central fatty marrow.

CT findings of the previously reported cases of extramandibular COF included expansile lesion with discrete areas of calcification and ossification (1-6). The proportion of soft tissue and calcification and ossification was variable. They were of low signal intensity on T2- and proton density-weighted MR images (4, 6). After infusion of contrast material, COF showed uniform dense enhancement (3, 4, 6). In our case, CT scans showed a large lobulated bone-density mass with a scanty soft-tissue component. MR imaging showed heterogeneous low signal intensity on T1- and T2-weighted images. After infusion of the contrast material, subtle enhancement was seen in a portion of the mass. These different CT and MR findings in our case might be due to increased amount of ossification.

It is not easy to differentiate mandibular COF from fibrous dysplasia. When it presents as a large lobulated bone-density mass in the parapharyngeal and masticator space, extrasosseous osteosarcoma, chondrosarcoma, and foreign-body injection granuloma should be differentiated.

Treatment should be directed toward complete removal of the mass, using enucleation or surgical resection with bone grafting. The prognosis is known to be fair, and recurrence after surgical removal seems to be unusual. Radiotherapy is contraindicated because of its radioresistance and post-radiation complications.

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