

A case report of cemento-ossifying fibroma presenting as a mass of the ethmoid sinus

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Abstract

Cemento-ossifying fibroma is a lesion containing both fibrous and osseous components. Such lesions include fibrous dysplasia, ossifying fibroma, cemento-ossifying fibroma and cementifying fibroma. Periodontal membrane is the origin of fibro-osseous lesions other than fibrous dysplasia.

Here a clinical case of a young woman referred for evaluation of a mass in the right side of face between eye and nose is presented. The first time she noticed the mass was 2 years ago and was growing larger inwards. She was treated with surgical resection.

In this case of a cemento-ossifying fibroma, histological interpretation was critical, and was the basis of correct treatment.

KEYWORDS: Fibroma, Ossifying, Ethmoid Sinus, Tomography, X-Ray Computed, Magnetic Resonance Imaging.

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Cemento-ossifying fibroma is considered to be a benign osseous tumor, very closely related to other lesions such as fibrous dysplasia; forming its own entity according to WHO classification in 1992.¹ It can be a bony tumor of possible odontogenic origin. It is believed to derive from the cells of the periodontal ligament.^{2,4} Differential diagnosis should be performed, preferably with other fibro-osseous lesions such as fibrous dysplasia or osseo-cementifying dysplasia.^{5,6}

It contains multipotential cells capable of forming cementum, lamellar bone and fibrous tissue. Under pathological conditions neoplasms contain any or all of the components that might be produced.⁷ They manifest themselves

as slow-growing, asymptomatic, intraosseous masses, most frequent in females aged between 35 and 40 years.^{8,9}

Case Report

A 27-year-old woman was referred for evaluation of a mass in the right side of face between eye and nose. The patient stated that the mass had first appeared 2 years ago and had been gradually increasing in size ever since. The host had non-specific headache, and no visual disturbances, dysphagia or dyspnea. Her medical history was normal. The physical examination revealed ethmoid area enlargement. Oral mucosa was normal.

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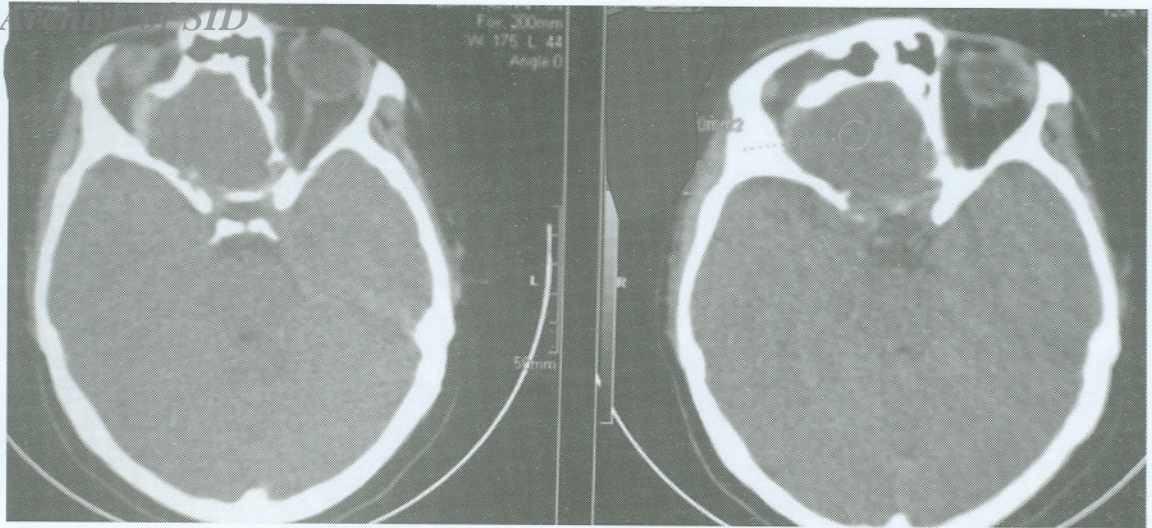


Figure 1. Brain coronal CT scanning

There is a soft tissue mass occupying right ethmoid and sphenoid sinuses destructing the bony lamina papyracea and minimally extended into the right globe. There is also destruction of the posterior wall of the mentioned sinuses and is extended into the right frontal lobe extra-axially.

On brain coronal CT scanning, there was a soft tissue mass occupying right ethmoid and sphenoid sinuses destructing the bony lamina papyracea and minimally extended into the right globe. There is also destruction of the

posterior wall of the mentioned sinuses and is extended into the right frontal lobe extra-axially (Figure 1). Areas of coarse curvilinear calcifications in the periphery of the lesion are noted.

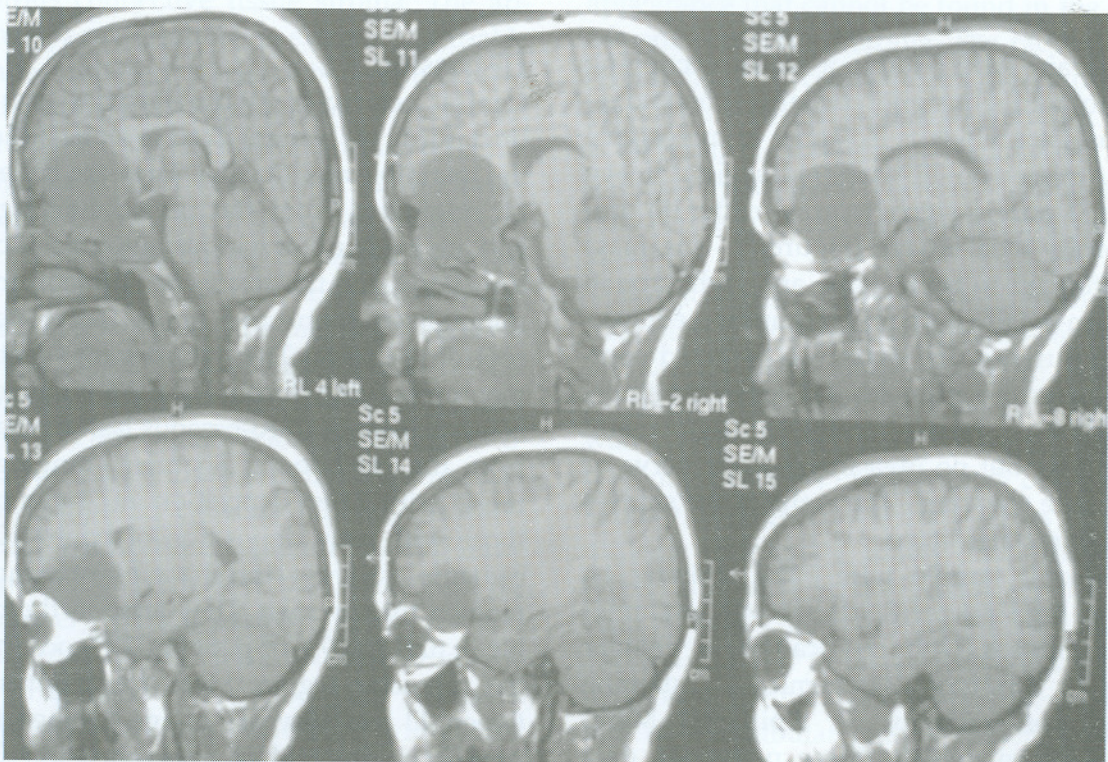


Figure 2. Brain MRI study

A mass occupying right ethmoid sinus and partially sphenoid sinus is seen which is isosignal to hyposignal to gray matter on T1-weighted sagittal image. www.SID.ir

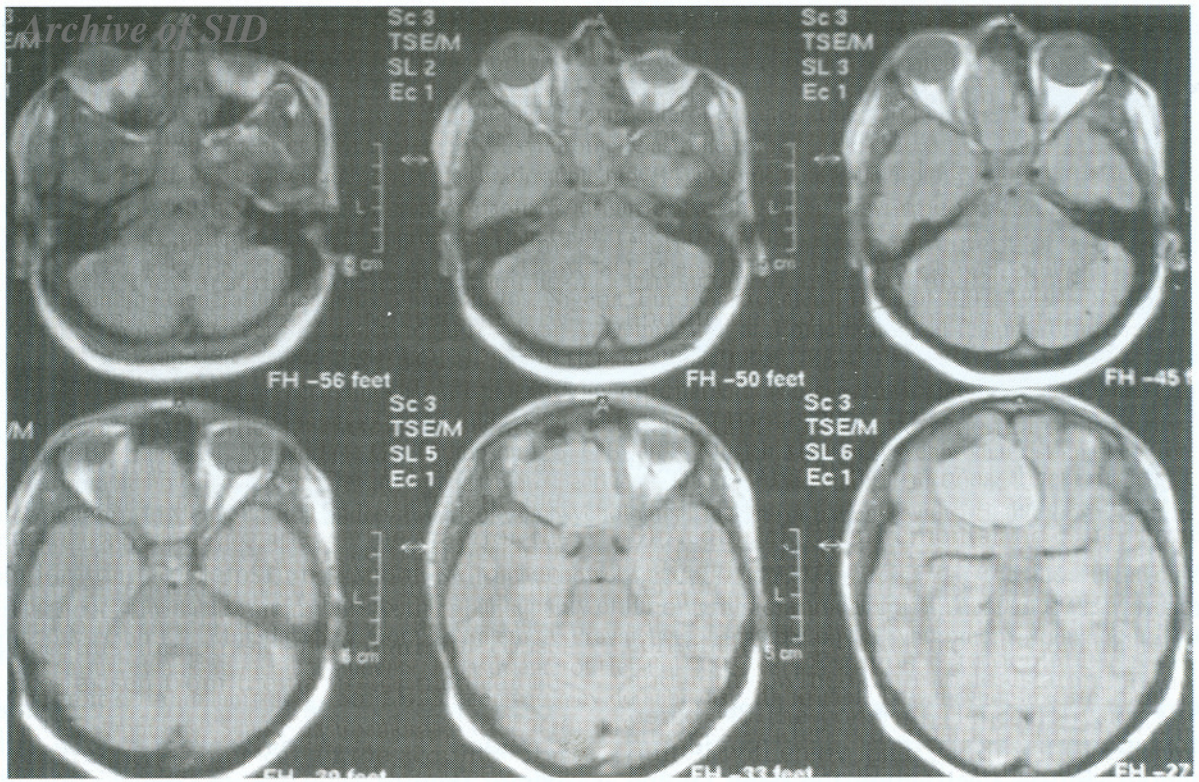


Figure 3. Brain MRI study

A mass occupying right ethmoid sinus and partially sphenoid sinus is seen which is isosignal to gray matter on proton density axial image.

On brain MRI study, a mass occupying right ethmoid sinus and partially sphenoid sinus was seen which is isosignal to hyposignal to gray matter on T1-weighted sagittal image (Figure 2), and isosignal to gray matter on proton density axial image (Figure 3). The mass was extended to right globe and right frontal lobe extra-axially with no surrounding edema.

After surgery, the mass was resected completely from the ethmoid and sphenoid sinuses. There was pressure on the right globe and frontal lobe, but no invasion to optic nerve or brain tissue was seen. The walls of the sinuses were also reconstructed.

Pathology sample with H&E staining on low and high power fields showed psammoma-like masses with ossification in cellular stroma. Immunohistochemical staining showed non-reactivity for Epithelial Membrane Antigen (EMA). These pathology data were compatible with a cemento-ossifying fibroma.

Discussion

Central cemento-ossifying fibromas are a distinct form of benign fibro-osseous lesions of the mandible and maxilla. They are believed to be originated from the periodontal ligament and are made up of various amounts of cementum, bone, and fibrous tissue. Cementum is the mineralized connective tissue that covers the root of the teeth. The hybrid name central cemento-ossifying fibroma is used because there is a spectrum of fibro-osseous lesions that arise from the periodontal ligament, ranging from those with only deposition of cementum to those with only deposition of bone.¹⁰ Central cemento-ossifying fibroma is more common in women than 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.¹⁰ When the tumor occurs in children, it is called the juvenile aggressive cemento-ossifying fibroma, which presents at childhood age and clinically

more aggressive and pathologically more vascular.¹⁰ Central cemento-ossifying fibromas are asymptomatic if they are not expanded. Therefore, they are generally not diagnosed until the tumor has had time to produce calcifications. Although the mandible is a common site for central cemento-ossifying fibromas, the maxillary sinus is an unusual site for central cemento-ossifying fibromas. Central cemento-ossifying fibromas are typically well-circumscribed, solitary radiolucencies with scattered radiopaque foci. They maintain a round shape, expand the surrounding cortical bone with no cortical violation, and may cause tooth divergence. The expanded tumors may involve the nasal septum, infraorbital foramen, and orbital floor. The extent of the tumor leads to surgical resection. At the time of diagnosis, maxillary central cemento-ossifying fibromas are large; indicating the ability of the tumor to expand freely within the maxillary sinus. Pathologic examination of the central cemento-ossifying fibroma represents a sum of irregularly-shaped calcifications within a hypercellular fibrous connective tissue stroma. The appearance of calcifications is extremely variable and shows different stages of bone and cementum deposition. Differentiation between cementum and osteoid is histologically difficult. In some cases, most of the calcified fragments are immature cementum, with basophilic coloration on hemotoxillin and eosin-stained sections, which are called central cementifying fibroma. In other cases, the calcified fragments are osteoid, with typical eosinophilic coloration on hemotoxillin and eosin-stained sections, which are called central ossifying fibromas. However, central ossifying fibromas can also be basophilic, causing difficulty in differentiating from central cementifying fibromas. Most pathologists believe that these two tumors namely central ossifying fibromas and central cementifying fibromas arise from the same progenitor cell; producing different amounts of bone and cementum within any one lesion. The hybrid central cemento-ossifying fibroma has evolved to indicate the likely presence of both types of tissue within

the same lesion. It is due to two reasons: difficulty in distinguishing reliably immature cementum from immature bone and the presence of both of these substances in many lesions. Therefore, the most accurate histological term is central cemento-ossifying fibroma, but it is interchangeable with either central ossifying fibroma or central cementifying fibroma. Apparently, there is no clinical or radiologic difference between the central ossifying fibroma and central cementifying fibroma, so the hybrid central cemento-ossifying fibroma is useful enough for radiology, too.¹⁰ The differential diagnoses includes other lesions that contain radiopacities within a rounded radiolucent mass: osteosarcoma or chondrosarcoma, odontogenic cysts, fibrous dysplasia, calcifying odontogenic cysts (Gorlin cysts), calcifying epithelial odontogenic tumors (Pindborg tumors), and squamous cell carcinomas. The factor that can differentiate central cemento-ossifying fibroma from aggressive sarcomas and carcinomas is its well-defined border. Fibrous dysplasia contains a characteristic of "ground glass" appearance that cannot be found in the central cemento-ossifying fibroma. The radiologic differentiation of central cemento-ossifying fibroma from Gorlin cysts and Pindborg tumors is difficult; the final diagnosis is based on histological appearance. There is a strong association between Pindborg tumors and impacted teeth.¹⁰

Surgical excision is the recommended treatment for the central cemento-ossifying fibroma. The entire tumor should be excised including involved regions of the maxillary sinus walls and orbital floor. Central cemento-ossifying fibromas usually "shell out" easily at surgery; however, maxillary types are more difficult to remove totally than mandibular types. 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. The recurrence rate of mandibular central cemento-ossifying fibromas is as many as 28% of patients. The recurrence rate of maxillary central cemento-ossifying 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.^{10,11}

Conflict of Interests

Authors have no conflict of interests.

Authors' Contributions

AH gathered radiology information. AGh gathered imaging information and wrote the case report. MS gathered surgery information. PM gathered pathology information. NT helped in writing the case report. All authors have read and approved the content of the manuscript.

References

1. Kramer IRH, Pindborg JJ, Shear M. Neoplasm and other lesions related to bone. In: Histological typing of odontogenic tumors. 2nd ed. Berlin: Springer;1992. p. 28-31.
2. Cohn HC, MacPherson TA, Barnes L, Kennerdell JS. Cemento-ossifying fibroma of the ethmoidal sinus manifesting as proptosis. *Ann Ophthalmol* 1982;14(2):173-5.
3. Fujimoto Y, Katoh M, Miyata M, Kawai T, Saito K, Morita M. Cystic cemento-ossifying fibroma of the ethmoidal cells (a case report). *J Laryngol Otol* 1987;101(9):946-52.
4. Brademann G, Werner JA, Jänig U, Mehdorn HM, Rudert H. Cemento-ossifying of the premaxillary region: case report and review of the literature. *J Laryngol Otol* 1997;111(2):152-5.
5. Su L, Weathers DR, Waldron CA. Distinguishing features of focal cemento-osseous dysplasia and cemento-ossifying fibromas: I. A pathologic spectrum of 316 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1997;84(3):301-9.
6. Su L, Weathers DR, Waldron CA. Distinguishing features of focal cemento-ossifying dysplasia and cemento-ossifying fibromas: II. A clinical and radiologic spectrum of 316 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1997;84(5):540-9.
7. Magu S, Airon RK, Mishra DS, Yadav RK, Kakkar V. Images: cementoossifying fibroma of the maxilla. *Ind J Radiol Imag* 2000;10(2):103-4.
8. El-Mofty SK. Cemento-ossifying fibroma and benign cementoblastoma. *Semin Diagn Pathol* 1999;16(4):302-7.
9. MacDonald-Jankowski DS. Cemento-ossifying fibromas in the jaws of Hong Kong Chinese. *Dentomaxillofac Radiol* 1998;27(5):298-304.
10. Kuta AJ, Worley CM, Kaugars GE. Central cementoossifying fibroma of the maxillary sinus: a review of six cases. *AJNR Am J Neuroradiol* 1995;16(6):1282-6.
11. Sanchis JM, Peñarrocha M, Balaguer JM, Camacho F. Cemento-ossifying mandibular fibroma: a presentation of two cases and review of the literature. *Med Oral* 2004;9(1):69-73. (English, Spanish)