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Tooth in the nose: Unusual presentation of a misdiagnosed maxillary ossifying fibroma

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Abstract

Fibro-osseous lesions involving the paranasal sinuses are uncommon, and making a determination of their histologic type is challenging. Correlating clinical and surgical findings with radiologic and histologic characteristics is essential for making a definitive diagnosis. We describe a case of misdiagnosed maxillary ossifying fibroma presenting as a solitary mass that completely filled and enlarged the left maxillary sinus. Another interesting finding was the presence of a molar tooth embedded in the tumor. We briefly comment on the genesis, natural course, and differential diagnosis of this lesion.

Introduction

The term *fibro-osseous lesion* is a descriptive but diagnostically imprecise term that applies to a group of nonspecific tumors in which normal bone is replaced by collagen, fibroblasts, and varying amounts of osteoid or calcified tissue.[1] The two most common types of fibro-osseous lesions are *fibrous dysplasias* and *ossifying fibromas,* which have similar histologic characteristics.[1-3] One of the confusing areas of pathology involves the examination of benign fibro-osseous lesions.[4] Making a careful differential diagnosis within this group of lesions is crucial because of the differences in their biologic behavior, treatment, and prognosis.[1,4]

Fibro-osseous lesions originate frequently in the bones of the extremities; they rarely occur in the craniomaxillofacial bones.[1] When ossifying fibromas originate in the craniomaxillofacial bones, they usually occur in the mandible (>70% of cases); other reported sites include the maxillofacial area and the paranasal sinuses.[2,5-7]

In this article, we describe an unusual presentation of a misdiagnosed maxillary ossifying fibroma, and we comment on the natural course and differential diagnosis of this lesion in light of our findings and in the context of the literature on this subject.

Case report

A 41-year-old woman presented to us with a progressive left-sided nasal obstruction and swelling of the left maxilla. She also complained of long-standing left-sided headaches and toothaches. She had been previously examined and treated by several different medical and dental practitioners. She had a history of tooth extractions and some dental surgical interventions, but little reliable documentation; no practitioners' notes were available for our review. The patient did possess a panoramic x-ray of her jaws that had been obtained 2 years earlier. Her medical history was otherwise unremarkable, and her family history was noncontributory.

On anterior rhinoscopy, a prominent nasal septal deviation to the left and mucopurulent nasal secretions were seen. Endoscopic nasal examination detected a white mass protruding from the medial wall of the maxillary sinus into the nasal cavity. Further examination was not possible because of the septal deviation. Examination of the 2-year-old panoramic dental x-ray revealed an abnormally located tooth-like opacity in the superior portion of the left maxillary sinus, as well as root resorption in many teeth, a periapical radiolucency on the first left upper molar, marginal periodontal bone resorption, and osseous lesions on the floor of the sinus (figure 1).

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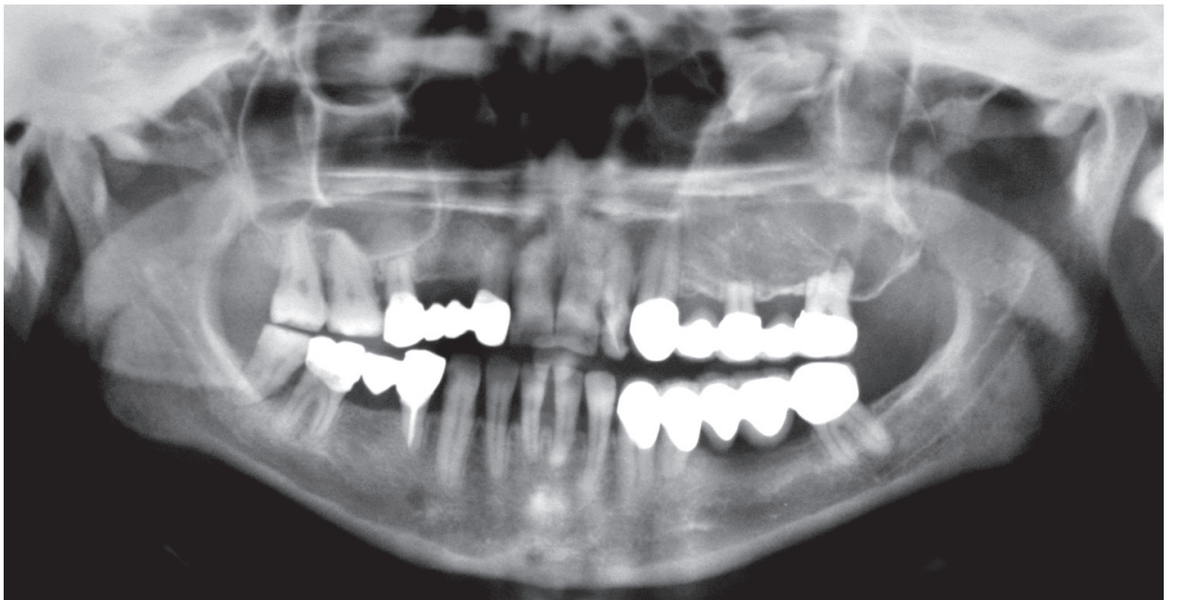


Figure 1. Panoramic x-ray obtained 2 years before our diagnosis shows the tooth-like opacity in the superior portion of the left maxillary sinus, as well as root resorption in many teeth, marginal periodontal bone resorption, and osseous lesions on the floor of the sinus.

Computed tomography (CT) of the paranasal sinuses demonstrated a well-demarcated solid mass consistent with a fibro-osseous lesion that had completely filled and enlarged the left maxillary sinus, as well as the presence of a molar tooth embedded in the tumor (figure 2). The tooth had protruded into the nasal cavity, just as had been seen earlier on nasal endoscopy.

Based on the clinical and radiologic findings, we reached a tentative diagnosis of a fibro-osseous lesion. Endoscopic endonasal surgery with intraoperative histologic investigation was planned. Following a nasal septoplasty, the crown of the displaced tooth was seen endoscopically. The tooth was located in the medial wall of the left maxillary sinus at a point posterior to the natural ostium of the sinus. Frozen-section histologic analysis identified the lesion as benign, and endoscopic medial maxillectomy was begun.

The tooth was extracted from the tumor to create more space for endoscopic instrumentation. However, we found it impossible to remove the tumor via an endoscopic approach because of its firmness and size. Therefore, a 4 × 4-cm bone window was opened on the anterior wall of the maxillary sinus via a gingivobuccal incision, and the tumor was removed via this combined approach.

The maxillary sinus was examined endoscopically, and no residual lesion was seen. Bony fractures had developed on the different walls of the maxillary sinus, but the orbital floor remained intact. No reconstruction was done for the defect on the anterior wall of the sinus.

The histologic examination revealed an ossifying fibroma characterized by the presence of acellular calcified spherules admixed with a moderately cellular spindle-cell stroma (figure 3). No osteoblastic rimming around the calcified spherules was present, and no new bone formation was seen. Mitotic figures were not found in the fibrous stroma. The histologic diagnosis was supported by negative results with immunohistochemical markers, including desmin, smooth-muscle actin, CD34, a cytokeratin AE1/AE3 cocktail, epithelial membrane antigen, glial fibrillary acidic protein, and Ki-67.

The postoperative period was uneventful. At the 2-month postoperative visit, the patient's toothache and nasal obstruction were persistent but less severe. No evidence of recurrence and no significant deformity of the facial contours were observed. No symptom of maxillary neuropathy was noted. The risk of recurrence and the need for frequent visits were discussed with the patient, but she was subsequently lost to follow-up.

Discussion

The differences between the two most common types of fibro-osseous lesions—fibrous dysplasias and ossifying fibromas—are well accepted in the literature.[4] Ossifying fibromas are true neoplasms that are confined to the jaws and craniofacial bones.[4,6] Brannon and Fowler have

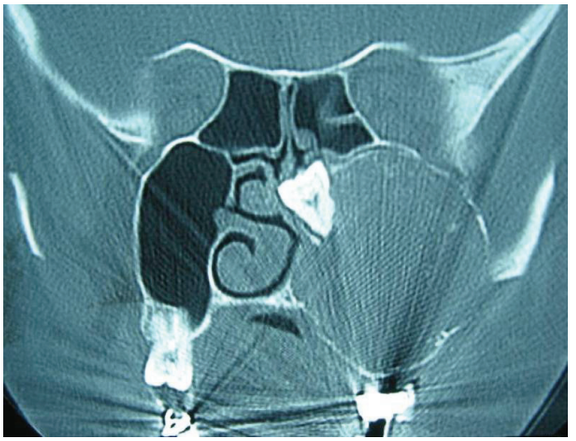


Figure 2. Coronal CT shows the tooth within the tumor involving the left maxillary sinus.

described the key diagnostic characteristics.[4] According to them, an ossifying fibroma:

- is a radiologically well-demarcated mass with smooth, often sclerotic borders;
- is a solitary lesion, usually located in the mandible;
- exhibits a centrifugal growth pattern (round/oval shape with enlargement);
- "shells out" from surrounding bone intact or in large pieces;
- has a vascular and cellular fibrous stroma; and
- features retiform bone trabeculae, some with osteoblastic rimming.

Additionally, when an ossifying fibroma arises in the jaw, the most common site of origin is the molar area.[4] Ossifying fibromas may absorb tooth roots, which may be an early finding if it can be detected on x-ray or CT,[7] as occurred in our case.

Brannon and Fowler have mentioned that correlation of the clinical and surgical findings with radiologic and histologic characteristics is crucial for a definitive diagnosis, as well as to avoid a late diagnosis.[4] As a self-criticism with respect to our case, we believe that it would have been more appropriate if we had performed a biopsy rather than an intraoperative histologic examination.

Treatment modalities for ossifying fibromas vary from watchful waiting to radical resection.[8] Complete surgical resection is the recommended treatment for craniomaxillofacial ossifying fibromas because of their locally aggressive nature.[1-8] Most ossifying fibromas are slowly growing and asymptomatic. If a definitive diagnosis is delayed, surgical excision may become more difficult. Therefore, ideal management should involve an early diagnosis and enucleation of the tumor at an early stage.[2]

The maxillary ossifying fibroma described in this report may be considered a slowly growing lesion in view of our patient's medical history. None of her other physicians had noted any abnormality related to an ossifying fibroma, and therefore no early diagnosis was made.

Surgical resection was more difficult than we expected because of the firmness of the tumor and its proximity to the sinus walls. We used a combination of endonasal endoscopic and gingivobuccal approaches for tumor resection. However, a marginal gingival incision would have been a better means of reaching the part of the lesion between the dental roots and for extracting the teeth if necessary. As illustrated by our case, a complete resection of these lesions via an endoscopic approach is not always possible. We felt that the patient's consent for a more radical surgical intervention should have been obtained, even though we had already planned an endoscopic surgery.

Ossifying fibromas continue to enlarge after normal bone growth is completed. Affected patients usually present between the second and fourth decades of life, and the female-to-male ratio is 5:1.[3,6] The biologic behavior and growth rate of ossifying fibromas are unpredictable; in addition to the typical slowly growing tumors, locally aggressive juvenile ossifying fibromas have been reported in the literature.[2,4,5] Reported recurrence rates have ranged between 1 and 63%, and malignant transformation is extremely rare.[3,4]

The genesis of ossifying fibroma of the mandible has been reported to be associated with molar development.[9] During this genesis, molars may be either located in

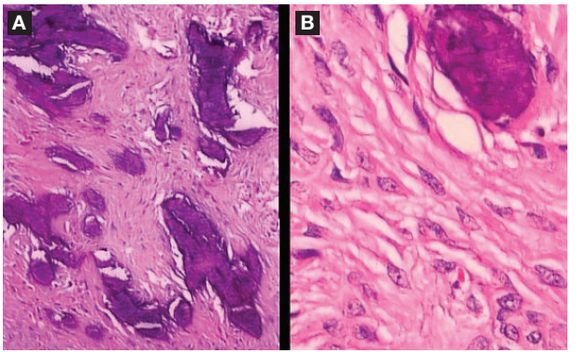


*Figure 3. Low- (**A**) and high-power (**B**) views show the acellular calcified spherules embedded in the cellular fibrous stroma (H&E, original magnification ×200 and ×1,000).*

their correct position, dislocated, or even missing entirely. In our patient, a molar tooth was displaced from its normal position. We believe that our case adds some weight to the argument in favor of an association between the genesis of maxillary ossifying fibroma and molar development.

Because our patient had long-standing dental problems and toothaches in addition to a tooth within her tumor, we propose a hypothesis related to the tumor's natural course. We hypothesize that this tumor most likely began to grow when the patient was about 18 to 20 years of age. As the tumor arose from the molar region, it carried the embedded third molar from its original location to the nasal cavity. We consider that this hypothesis may suggest the slowly growing behavior of this ossifying fibroma from youth to middle age. Perhaps our patient's tumor was a long-unrecognized juvenile ossifying fibroma. Beyond these speculations, further interpretation of a solitary case is not meaningful.

In conclusion, we describe an unusual presentation of a maxillary ossifying fibroma. Although our report involves only a single case, we believe that it is of value because the available data on the genesis and natural course of maxillofacial ossifying fibromas are very limited. We believe that this report provides useful information to otolaryngologists and oral maxillofacial surgeons, as well as dental practitioners, regarding the scope of maxillofacial fibro-osseous lesions.

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