CLINICOPATHOLOGIC CONFERENCE

A large painless swelling of the posterior mandible

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CLINICAL PRESENTATION

A 60-year-old man presented with primary complaint of a large painless swelling of the left side of the mandible. On examination, an obvious swelling was present on the left side of the body of the mandible, extending anteriorly from the left mandibular first premolar to the angle of the mandible posteriorly; involving the lower border of the mandible, causing its expansion and thinning and also showing buccolingual cortical expansion. The patient's medical history was noncontributory, and physical examination revealed no other abnormality. The lesion was covered by normal-appearing mucosa. A panoramic radiograph revealed the presence of an ill-defined, multilocular radiolucent lesion in the body of mandible (Figure 1).

DIFFERENTIAL DIAGNOSIS

Based on the ill-defined, multilocular radiolucent presentation of this large expansile mass of the posterior mandible on orthopantomograph, the differential diagnosis for this lesion included benign locally aggressive odontogenic tumors, including ameloblastoma, odontogenic myxoma, and keratocystic odontogenic tumor; central giant cell granuloma; aneurysmal bone cyst; central AV malformations; and metastatic carcinoma.

Ameloblastoma is the commonest benign tumor of odontogenic origin that develops from remnants of odontogenic epithelium, more specifically from the rests of the dental lamina. It is generally a slow-growing but locally invasive tumor. The peak incidence is in the third to fourth decades of life, with most cases occurring in the posterior mandibular segment. In the present case, however, the patient's age was 60 years, which was not in favor of ameloblastoma. Painless cortical expansion of the affected jaw causing facial deformity and loosening of teeth is the most common presenting sign. This neoplasm presents radiographically as a well-defined unilocular-to-multilocular radiolucency, giving the lesion a "honeycomb" or "soap-bubble" appearance.[1,2] Even though the fine soap-bubble appearance was absent in the present case, the multilocularity and the site of the lesion suggested ameloblastoma.

Odontogenic myxoma, a rare benign odontogenic tumor, develops from odontogenic ectomesenchyme of a developing tooth or undifferentiated mesenchymal cells in the periodontal ligament, and is locally invasive.[1] It is generally depicted as a slow-growing tumor with the potential to attain considerable size without noticeable signs and symptoms. It frequently displays aggressive infiltration of the adjacent tissue, as well as a tendency to recur after surgical removal. It is most frequently seen in patients in the second to fourth decades of life, with molar and ramus regions of the mandible as the most frequent sites of occurrence. Radiographically, it may have many presentations: unilocular or multilocular; well-defined, corticated; well-defined, noncorticated; and poorly defined or diffuse lesion.[2,3] Odontogenic myxomas can be extensive, with thinned cortex owing to expansion, and may perforate at a later stage of tumor progression.[3,4] The wispy trabeculae typically seen in odontogenic myxoma were not seen in the radiograph of the present case, but the aggressive behavior leading to bony expansion and the irregular radio-

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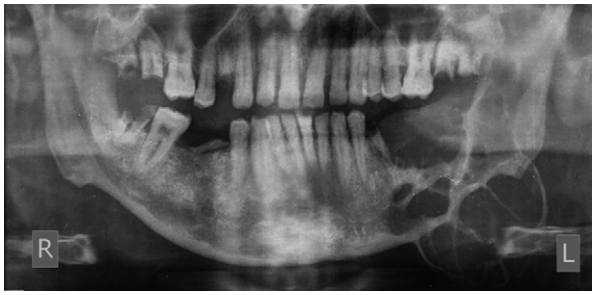

Fig. 1. Panoramic radiograph showing an ill-defined, multilocular radiolucency of the left side of mandible.

graphic outline hinted toward an aggressive lesion, such as odontogenic myxoma.

Odontogenic keratocyst (OKC), referred to as "keratocystic odontogenic tumor" (KCOT) in the latest World Health Organization classification (2005), is an aggressive, benign intraosseous neoplasm of odontogenic origin with characteristic lining of parakeratinized squamous epithelium.[5] The peak incidence is in the second and fifth decades of life, with most cases occurring in the mandible, especially the posterior portion of the body.[5,6] Buccal expansion is noted in approximately 50% of the mandibular lesions. Clinical manifestations include swelling, pain, or both, but it may also be asymptomatic. Radiographically, it may appear as a unilocular, or more commonly, a multilocular, radiolucency with pronounced bone resorption.[6,7] Odontogenic keratocyst rarely shows a buccolingual expansion and usually presents as a well-defined radiolucency rather than the irregular radiographic picture of the present case.

Central giant cell granuloma of the jaws is usually a non-neoplastic bone lesion accounting for fewer than 7% of all benign tumors of the jaws. The clinical presentation varies from a slowly growing asymptomatic swelling to an aggressive lesion associated with pain, cortical bone destruction, root resorption, and displacement of the teeth. The peak incidence occurs in the second decade of life, with 71.8% of patients younger than 30 at the time of diagnosis, with a female predilection. The anterior mandible is the most common site of occurrence, often crossing the midline. Radiologically, the lesion presents as a well-defined, unilocular or multilocular radiolucency, with varying degrees of expansion of the cortical plate.[8] Central giant cell granuloma was considered here as a differential diagnosis because of the aggressive nature of the lesion and the buccolingual expansion, in addition to the multilocularity seen radiographically, despite the age of the patient.

Aneurysmal bone cyst (ABC) is an infrequent bone lesion, presenting as less than 1% of all bone cysts.[9] Although ABC is a benign lesion, it can behave locally in an aggressive manner because of its rapid growth and osteolytic capacity.[5] The patient presents commonly with dull pain and/or edema. Radiographic findings usually include unicystic or, occasionally, multilocular bone expansion, or having a honeycomb or soap-bubble–like structure, sometimes with destruction of the bony cortex and periosteal reaction.[9,10] Unlike the current case, ABCs are usually seen in individuals younger than 30 years and are associated with pain.

Central arteriovenous (AV) malformations are extremely rare conditions that are potentially lethal if left untreated, as a result of massive blood loss after extraction or attempts to remove or biopsy the lesion.[11] AV malformations represent anomalous blood vessels where the walls of the arteries and the veins directly interconnect via multiple abnormal vascular routes. High arterial pressure is exerted within the vessels because of direct communication of arteries with veins. This often causes compensatory expansion of cortices, and a bruit can be heard on auscultation. Radiographically, it most commonly appears as a multilocular radiolucency. Biopsy should not be performed if on aspiration bright red fresh blood is obtained.[12]

Metastatic tumors to the jaws are rare lesions that often present with innocuous symptoms mimicking dental infection. Theoretically, any malignant tumor can metastasize to the head and neck area. The angle and body of the mandible are the most favored sites of deposition, probably because of the rich intraosseous vascularity in these areas. The most important clinical findings are swelling, paresthesia of lip, and deep bone pain. Radiographically, these present as irregular, moth-eaten expansile radiolucencies. Despite the absence of any history of malignancy, the possibility of metastatic disease should be considered because, most of the time, patients may not be aware of an underlying primary malignancy.[13,14]

DIAGNOSIS

Aspiration with respect to the lesion was nonproductive, thus excluding the possibility of vascular and cystic lesions. Taking into account the size and the aggressive nature of the lesion, a working diagnosis of ameloblastoma or central giant cell granuloma was made. An incisional biopsy was thus performed under local anesthesia, and the specimen was submitted for histopathological examination.

Microscopic examination of the specimen revealed sheets of malignant cells resembling plasma cells. Most of the malignant cells were round to oval, with eccentrically placed hyperchromatic nuclei exhibiting chromatin clumping ("cartwheel" appearance), with a few cells showing different degrees of maturity, ranging from un-