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## CLINICAL PATHOLOGIC CONFERENCE CASE 3: A CHALLENGING CASE OF AN ENLARGING SWELLING OF THE MAXILLA

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**Clinical Presentation:** A 32-year-old Nigerian female presented to a local dentist with a 2-month history of an enlarging swelling of the upper left labial sulcus, extending from the central incisor to the canine area. The patient had never attended a dentist before this.

Panoramic tomography (Figure 1) showed a radiolucent lesion in the anterior aspect of the left maxilla at the level of the alveolus. This lesion was predominantly well-circumscribed, but on the right side, close to the apices of the incisors and canine, the borders were indistinct. The lesion also appeared to be extending into the right side and midline of the nasal septum. Although predominantly osteolytic, there were a few sclerotic foci toward the left side. The hard palate appeared thinned but not resorbed. The lesion was not corticated and there was marked irregular erosion of the tooth roots of the first and second incisors and of the apical area of the canine (Figure 1, inset).

**Differential Diagnosis:** Although the clinical symptoms were present for only 2 months, the radiographic findings are compatible with a lesion of long duration, with a rather slow but progressive rate of growth. Therefore, the entities to be discussed in the differential diagnosis should include benign but locally aggressive tumors (odontogenic and non-odontogenic), malignant lesions (mainly of the low-grade type), and inflammatory/infectious conditions.

Among the benign odontogenic tumors, desmoplastic ameloblastoma (DA) can be considered because it manifests as a painless swelling of the jaw, usually located in the anterior region.[1] The slow rate of growth is consistent with movement of tooth roots and root resorption, as seen in this case. Radiographically, DA has ill-defined borders. Although our case is partly ill-defined and infiltrative, it is mostly well-circumscribed. Additionally, DA often has a mixed radiolucent-radiopaque pattern, similar to a fibro-osseous lesion, which was not seen in the present case.

Odontogenic fibroma (OF) and calcifying epithelial odontogenic tumor (CEOT) are also included in the differential diagnosis. OF has a female predominance and, when present in the

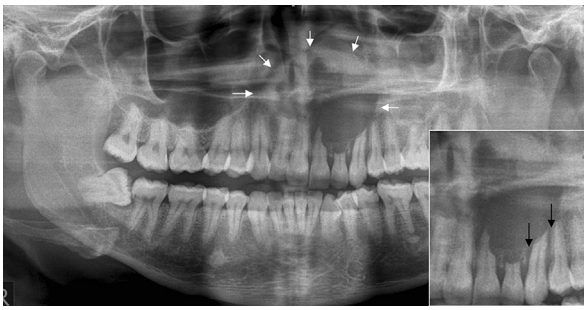

Fig. 1. Panoramic tomography shows an extensive radiolucent lesion that occupies the left maxilla and crosses the midline. Destruction of the left maxillary sinus is evident and the left hard palate is thinned but not resorbed. The white arrows mark the estimated borders of the lesion. This lesion is predominantly well-circumscribed but on the right side (close to the apices of the incisors and canine) the borders are indistinguishable. The lesion affects the adjacent dentition, which exhibits tilting, extrusion, and root resorption. Given the limits of this type of radiographic image, it seems that the dentition is not affected by caries or periodontal disease. **Inset**: Spiking root resorption of the right central incisor and right canine is remarkable. Widened periodontal ligament space is also observed (*arrows*).

maxilla, tends to affect the anterior portion.[2,3] However, CEOT is more commonly noted in the premolar-molar region of the mandible. Both OF and CEOT usually present as slowly enlarging painless expansion of the jaws that, on imaging, can present as radiolucent lesions with evidence of tooth displacement and resorption.[2-4] OF is often very well-demarcated or corticated; CEOT often presents as a mixed radiolucent-radiopaque lesion on imaging (Figure 1).

Among the malignant odontogenic tumors, ameloblastic carcinoma[5] and clear cell odontogenic carcinoma[6] may be considered. Ameloblastic carcinoma and clear cell odontogenic carcinoma can show an expansile intra-osseous radiolucency with ill-defined borders and root resorption. Perforation of the bone cortices and infiltration into adjacent structures can also be present. However, these lesions are mainly located in the mandible; additionally, pain and/or paraesthesia may also be expected.[3] None of the latter symptoms were present in our case.

The central giant cell lesion (CGCL) in most cases is asymptomatic and can show swelling and loosening of teeth in young adult patients (average age, 25 years). On imaging, CGCL presents as a multilocular or unilocular radiolucent lesion with non-corticated borders. Disappearance of the lamina dura, root resorption, tooth displacement, nasal obstruction, and penetration of the jaw cortex have been described in CGCL.[7,8] However, the majority of cases are located in the mandible.

Sinonasal schwannoma is a rare tumor constituting only ~4% of all head and neck schwannomas.[9] It can be painless, even when tumors are large and extend into adjacent structures.[9] In long-standing lesions, cystic change can occur and this may confer a radiolucent appearance in a background of increased radiopacity,[10] as in the present case. Tooth movement and root resorption have been documented in schwannomas of the jawbones.[11]

Sinonasal lymphoma is the second most common malignancy of the sinonasal tract, following squamous cell carcinoma, with B-cell lymphomas, being the more frequent type found in the paranasal sinuses.[12-14] The radiographic features usually demonstrated a poorly demarcated lesion with expansion into adjacent structures, bone destruction, and root resorption and widening of the periodontal ligament of adjacent teeth. Some of these features were evident in the present case. Similar radiographic features can be encountered in osteosarcoma of the jawbones.

Sinonasal squamous cell carcinoma[15] and sinonasal adenocarcinoma[16] are not favored diagnoses because they usually arise higher in the sinonasal region and are expected to be symptomatic, especially in advanced stages. The patient in the present case was asymptomatic, despite the advanced stage of the tumor.

Of the inflammatory/infectious conditions, rhinoscleroma could be considered because it is a chronic, progressive process with potential to destroy and extend into adjacent structures, including the oral cavity.[17] However, this is a less likely diagnosis in the present case, as the patient is not known to reside in an area endemic for rhinoscleroma.

Finally, metastasis to the jaws cannot be excluded. However, the current patient is relatively young to have metastatic disease. The majority of the patients with metastases to the oral cavity and jaws are older than 50 years of age.[18] Breast, prostate, and lung cancers are the most common primary tumors presenting with jaw metastasis.[18,19] Other primary sites of origin of metastases reported in the jaws are cancers of the colon, liver, rectum, thyroid, uterus, and parotid gland.[18,19]

**Diagnosis and Management:** The macroscopic features of the incisional biopsy consisted of irregular pieces of tan-colored tissue with a maximum dimension of 15 mm. Hematoxylin and eosin-stained sections showed a densely cellular spindle cell tumor (Figure 2) arranged in interlacing fascicles that in places resembled a herringbone pattern. Cytologically, the cells were fusiform and spindled with mild pleomorphism. Scattered within the lesion were infrequent small islands of irregular osteoid (Figure 2). Some of these osteoid structures were surrounded by large mildly pleomorphic cells consistent with osteoblasts.

The histopathologic differential diagnosis was broad and initially included tumors such as monophasic synovial sarcoma, fibrosarcoma, fibroblastic osteosarcoma, chondrosarcoma (there was no evidence of chondroid tissue), and phosphaturic mesenchymal tumor. However, given the areas of osteoid formation, the histopathology was most suggestive of a malignant mesenchymal tumor consistent with osteosarcoma. Immunohistochemistry for a broad sarcoma panel that included cytokeratins, EMA, S100, desmin, CD34, and CD31 was negative. Patchy positivity for SMA, CD99, and CD117 was seen. The Ki67 proliferation rate was about 15% of the tumor cells.

The case was referred for management to the Royal National Orthopaedic Hospital and the University College London Hospital. Both institutions agreed with the diagnosis of a fibroblastic osteosarcoma.

Following the diagnosis, computed tomography of the head and neck was obtained (Figure 3). This showed the main bulk of the tumor was in the left maxilla with extension across the midline to involve the right maxilla, upper lip, and premaxillary soft tissues. The lesion displayed irregular boundaries and contained small flecks of mineralization.

The patient was given preoperative chemotherapy followed by a subtotal maxillectomy (Figure 4) at the University College Hospital, London in 2009. The resection specimen included most of the left maxilla, anterior part of the right hard palate, upper lip and anterior part of nose.

The histology from the resection specimen showed features similar to the incisional biopsy. However, there were numerous markedly pleomorphic cells associated with areas of mineralized