A Rare Case of Odontoameloblastoma in the Geriatric Population

Pratyusha Yalamanchi BA[1], Orly Coblens MD[1], Meejin Ahn DO[2], Steven B. Cannady MD[1], Jason G. Newman MD[1]

[1]Department of Otorhinolaryngology - Head and Neck Surgery, Hospital of the University of Pennsylvania, West 34th Street, Philadelphia, PA 19107, USA [2]Department of Pathology, University of Pennsylvania Health System

Background

Odontoameloblastoma (OA) is an extremely rare tumor derived from odontogenic epithelium and mesenchyme. In the fewer than 20 reported cases that fulfill histological classification, OA is described as asymptomatic involvement of the mandible or maxilla in young men with a median age at diagnosis of 20 years and a history of unerupted teeth. The lesion is characterized by a slow, progressive growth pattern similar to other odontogenic tumors such as solid multi-cystic ameloblastoma. Wide local excision and persistent surveillance are recommended given the locally destructive growth pattern and recurrence risk.

Objective

Here, we present the first reported case of odontoameloblastoma in a patient over age 50. Though considered a disease of young people, OA should be included in the broader differential of lytic lesions involving the maxilla or mandible.

Case Report

A 73 year-old female presented to the dentist for routine cleaning and x-rays, which displayed a lesion involving the mandible. After referral to multiple providers, a biopsy of two involved teeth and CT neck were performed. Imaging and biopsy revealed a large, destructive lesion of the mandible with histology consistent with odontoameloblastoma. The patient underwent wide segmental mandibular resection and scapula tip free flap reconstruction. She recovered uneventfully and continues to have close follow-up given risk of recurrence.

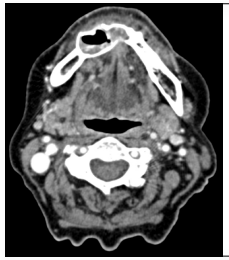

Figure 1. Axial cut of the CT scan revealing a 2.7cm x 3.5cm x 2.5cm destructive, loculated lesion involving the right anterior horizontal ramus of the mandible which extended across the midline and abutted the neural foramina. Gas density and cortical bone thinning was present along the margins.

Results

Gross Surgical Pathology and Histologic Findings

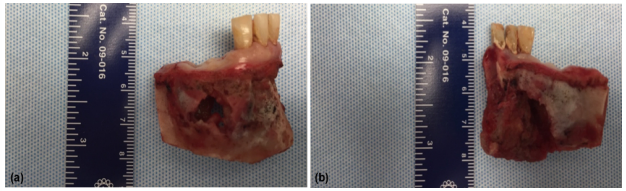

Figure 2. (a) Gross surgical pathology revealing portion of a partial right mandible with three attached teeth measuring 4.8 x 3.0 x 1.8 cm, anterior view (b) Posterior view of gross specimen

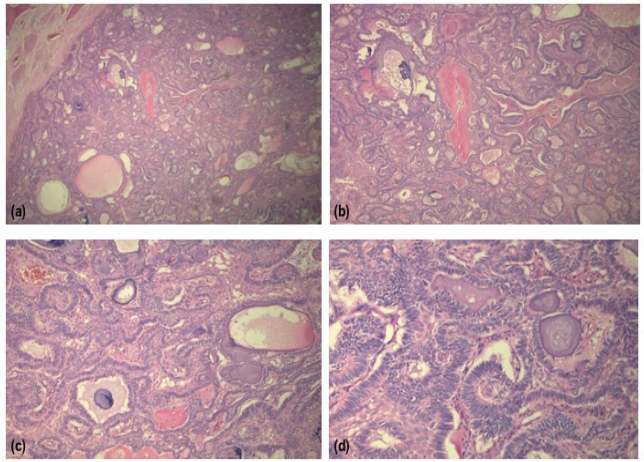

Figure 3. (a) Photomicrograph showing odontogenic epithelium adjacent to cellular myxoid tissue and foci of dentin-like material (H&E stain, ×2.5). (b) Photomicrograph showing variable amount of mature connective tissue adjacent to the epithelium with mineralized dental tissues (H&E stain, ×5). (c) Photomicrograph further depicting islands and cord of odontogenic epithelium with follicular and plexiform patterns surrounded by myxoid tissue and dentinoid enclosing pulp like space (H&E stain, ×10). (d) Photomicrograph displaying nuclear palisading and stellate reticulum seen with ameloblastoma (H&E stain, ×20)

Discussion

Presentation of OA and Differential Diagnosis

Despite asymptomatic presentation, OA is characterized in the literature as a progressively growing, centrally destructive lesion that leads to swelling of alveolar bone, malocclusion, and dull pain. Tumor growth can result in destruction of surrounding bony tissues, leading to possible obstruction of the nasal and oral airways if left untreated.

As in our case, OA classically appears on imaging as a well-defined, radiolucent lesion that may contain radiopaque particles, indicative of dentin, or a larger radiopaque central mass, similar to a complex odontoma. Differential diagnosis include compound or complex odontomas, calcifying epithelial odontogenic tumors, ameloblastic fibroodontomas, and cement-ossifying fibromas.

Histologic Diagnosis and Management

Histologic diagnosis is required to differentiate OA from other odontogenic tumors. OA lesions fulfill three histopathologic criteria, specifically presence of (1) ameloblastoma, (2) homogeneous, mature connective tissue, and (3) fragments of calcified dental structures. This combination of undifferentiated neoplastic tissue with highly differentiated tissue contributes to the unusual pathology of OA.

Given its potential for bone expansion and local destruction, OA is treated with wide excision and close surveillance for several years. A reported 21.4% rate of recurrence merit a minimum of five years of close follow-up. A multidisciplinary team, including primary care dentists and oral and head and neck surgeons, is often required to optimize care.

Conclusions

- The present case demonstrates the diagnosis and management of OA, in an older demographic than previously described.
- Though considered a disease of young men, OA is an important diagnostic consideration in all patients presenting with lytic lesions involving the mandible or maxilla, as the tumor is locally destructive and surgical resection is curative.

Penn Medicine