Odontoameloblastoma: Report of two cases

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ABSTRACT

Odontoameloblastoma (OA) is a very rare mixed odontogenic neoplasm, characterized by the simultaneous occurrence of an ameloblastoma and a compound or complex odontoma in the same tumor mass. To date, less than 50 cases of OA and/or ameloblastic odontoma have been reported in the English dental literature. This neoplasm was called ameloblastic odontoma. The term OA was included in the 1971 WHO classification. In this study, we present two cases of OA, which we hope will contribute to the awareness and knowledge of surgeons regarding the existence of this odontogenic tumor so that patients having it may be treated and followed-up properly.

Key words: Ameloblastoma, mixed odontogenic tumors, odontoameloblastoma, odontogenic tumors, odontoma

Odontoameloblastoma (OA) is an extremely rare odontogenic tumor that contains an ameloblastomatous component together with odontoma-like elements.[1] It is similar to an ameloblastoma, both in structure and in behavior.[2] This tumor was formerly called ameloblastic odontoma. Thoma *et al.*[3] described the first case in 1944 and since the first edition of the WHO Histological Classification of Odontogenic Tumours,[4,5] the OA still appears as a distinct odontogenic neoplasm.[6] To date, there are less than 50 cases that have been reported as OA and/or ameloblastic odontoma in the English dental literature.[7,8] The epithelial proliferation portion of the tumor forms islands or cords resembling the follicular or plexiform pattern typical of an ameloblastoma. This ameloblastic component is intermingled with dental tissues of variable degrees of maturity, as seen in odontomas.[1] We present two cases of OA with different clinical characteristics, except for the fact that both occurred in young patients.

CASE REPORTS

Case 1

The patient, a 22-year-old black woman was referred to the clinic because of a swelling located in the left upper area of the vestibule of the mouth. Extraoral examination revealed a discrete swelling of both the left upper lip and the nose wing [Figure 1a]. Intraorally, a painless swelling on the left side of the maxilla extending from the central incisor to the first premolar was observed [Figure 1b and 1c]. Even though these teeth were slightly mobile, they were still vital. The consistency of the swelling was uniformly hard and the overlying mucosa was purple. The past medical history of the patient was non-contributory. Radiographic examination included occlusal [Figure 2a] and periapical [Figure 2b] radiographs. A cyst-like intraosseous lesion was found with a well-defined margin. The lesion was located in the apical region of the left central incisor, lateral incisor, canine and first premolar. These teeth presented root resorption [Figure 2a and 2b]. The lesion, exhibiting different levels of radiolucence, did not cross to the right side of the maxilla. At the uppermost side of the lesion, a radiopaque mass with radiodensity similar to that of the dentine was present. An adenomatoid odontogenic tumor, a calcifying epithelial odontogenic tumor and an ameloblastic fibroodontoma were considered as differential diagnoses. An incisional biopsy was planned under local anesthesia; however, during the surgical procedure, the lesion was wholly detached, resulting in an excisional biopsy [Figure 1c]. The tumor was surgically enucleated with a 2mm safety margin and the material was fixed in a 10% formalin solution and submitted to histopathological analysis. Figure 3a-3c shows the histopathological findings for the tumor. Figure 3a reveals areas of plexiform ameloblastoma with some cystic spaces in the stroma. Epithelial proliferation in the form of anastomosing cords could be seen. Cubical to columnar cell-surrounded areas resembling the stellate reticulum of the enamel organ were seen. These areas were surrounded by a mesenchymal component [Figure 3b]. Calcified structures were observed

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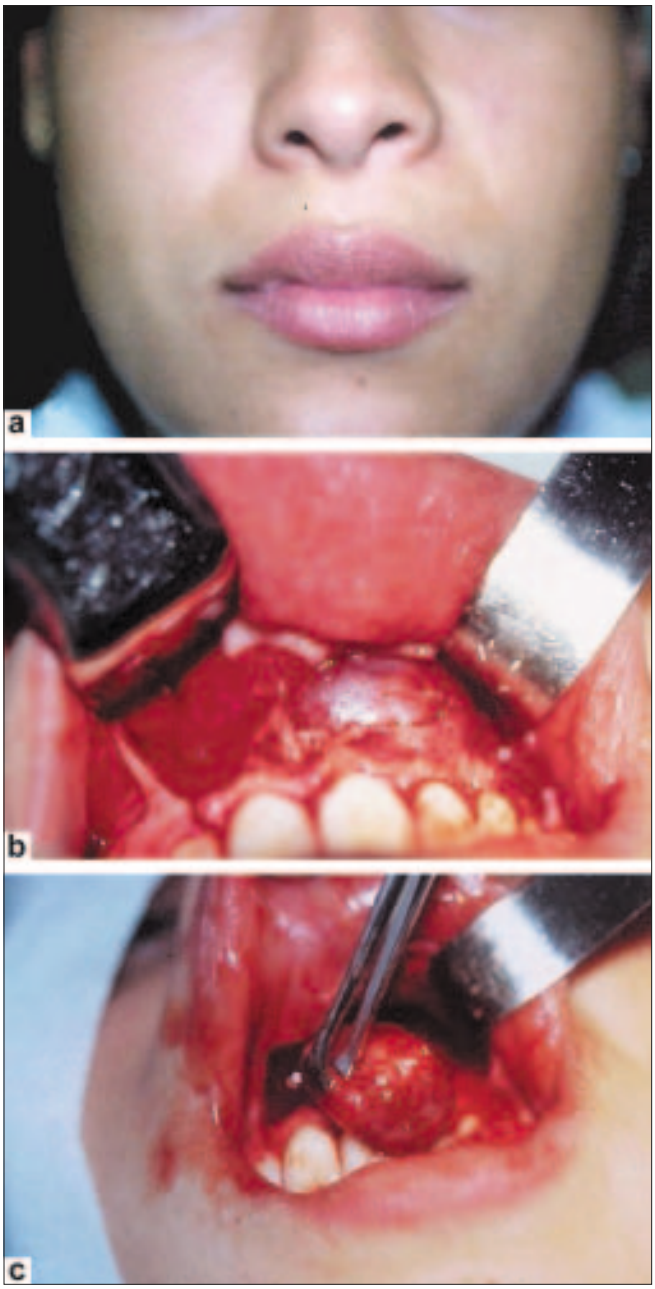

Figure 1: Clinical aspects of the lesion

in the mesenchymal component [Figure 3c]. These structures resembled immature dentine (eosinophilic structure) and cementum (basophilic structure) of variable degrees of maturity. Based on the radiographic and histopathological findings, a final diagnosis of OA was made. There was no recurrence of the tumor after a 6-year follow-up.

Case 2

The patient, a 16-year-old Caucasian man, was referred to the clinic because of a swelling located in the right mandibular molar region. Extraoral examination revealed a swelling of the posterior mandible, which was hard in consistency. The lesion was painless. It had been detected and was growing slowly for the past two and a half years. The medical histories of the patient and his family were non-contributory. Intraoral examination revealed a poorly circumscribed hard swelling in the molar area. The covering mucosa was normal in color and texture. Radiographic examination included posteroanterior [Figure 2c] and lateral [Figure 2d] radiographs of the mandible. In both radiographs, a large and irregular radiopaque mass was observed circumscribed by a radiolucent rim. The radiopaque mass had globules with different degrees of radioopacity. The lesion produced an expansion of the vestibular cortical bone [Figure 2c]. Based on the clinical and radiographic examination, a diagnostic hypothesis of odontoma was considered and an excisional biopsy was indicated. The lesion was removed under general anesthesia through an extraoral access. The material was fixed in a 10% formalin solution and submitted to histopathological analysis. Figure 3d–3f shows the histopathological findings for the tumor. Figure 3d reveals sheets of odontogenic epithelium resembling that of a solid ameloblastoma, surrounded by a mesenchymal component. Under higher magnification [Figure 3e], a proliferating odontogenic epithelium could be seen as sheets delimited by columnar epithelial cells in close contact with the mesenchymal surrounding tissue. Calcified structures/material was associated with the mesenchymal component [Figure 3f]. These structures had the morphological features of immature dentine (eosinophilic structure) and cementum (basophilic structure). Based on the radiographic and histopathological findings, a final diagnosis of OA was made. There was no clinical or radiographic evidence of recurrence of the tumor after a 6-month follow-up.

DISCUSSION

Because of the rarity of OAs, little reliable information is available. According to Mosqueda-Taylor *et al.*,[7] only 14 cases reported in the literature met the WHO histological and clinical criteria to be classified as OAs. In their review of the literature, these authors discussed all the misdiagnosed lesions that were confused with OAs, especially when ghost cells were present in the tumors.[9,10] The incidence of OAs is very low. In a review of 164 odontogenic tumors, Stypulkowska[11] found a single case of OA (0.6%). In another review of 108 odontogenic tumors previously diagnosed as ameloblastomas, Raubenheimer *et al.*,[12] also found a single case of OA (0.9%).

Radiographically, the tumor presents a radiolucent, destructive process that contains calcified structures resembling mature dental tissue. These radiographic features were seen in our cases.

OAs have clinical and microscopic characteristics that allow them to be differentiated from typical ameloblastomas and odontomas. They occur in young patients and have a predilection for males.[4,5] Pain, delayed eruption of teeth and expansion of the affected bone may be observed. As regards

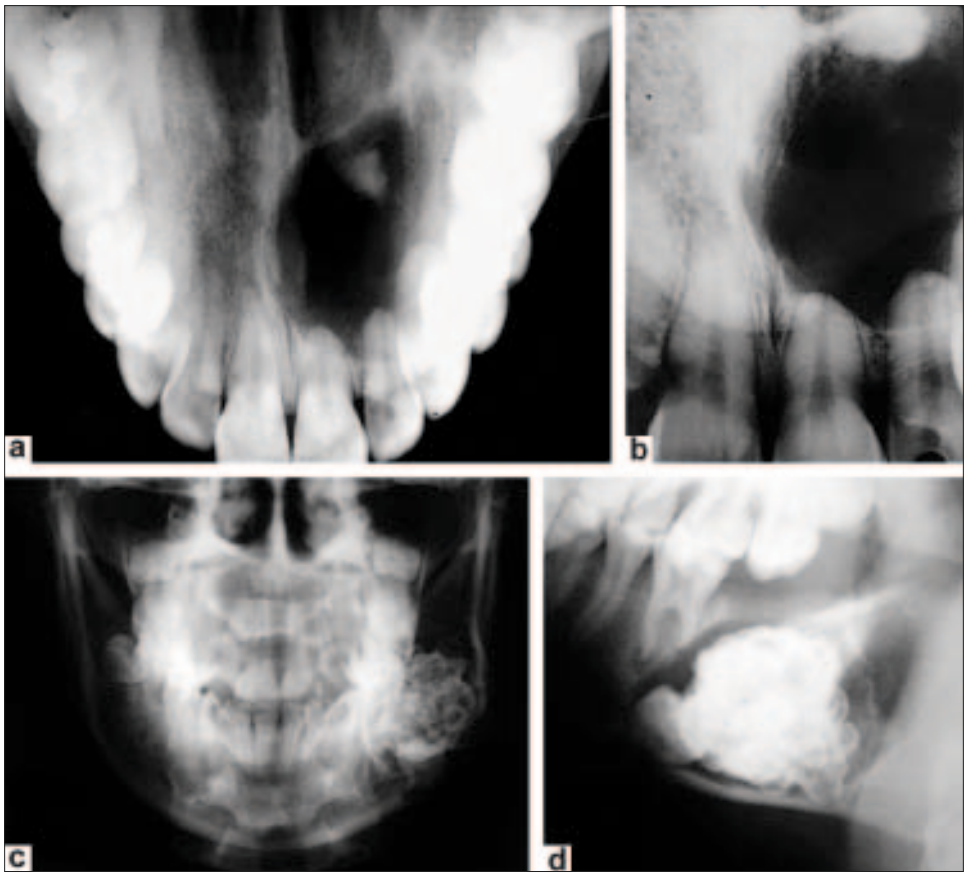

Figure 2: Radiographic examination. First case: (a) occlusal radiograph and (b) periapical radiograph. Second case: (c) posteroanterior and (d) lateral radiograph of the mandible

location, according to Mosqueda-Taylor *et al.*,[7] the tumor can affect both the maxilla and the mandible equally, whereas Martín-Granizo López *et al.*[8] found that there is a slight tendency toward greater involvement of the mandible.

Because OAs are rare, only case reports and literature reviews citing these cases are available, except for the study by Yamamoto *et al.*,[13] who studied the behavior of the tumor by analyzing proteins of the extracellular matrix of the basement membrane. No studies were found establishing the pathogenesis of this neoplasm. Nevertheless, some theoretical explanations were presented. According to Thompson *et al.*,[14] the proliferating epithelium could induce the mesenchymal tissue to form hamartomatous mineralized dental tissue. Mosqueda-Taylor *et al.*[7] presented another possibility: The coexistence of an ameloblastoma and an odontoma. They would develop separately and then collide. The same authors, however, discarded this explanation based on the clinical aspects of these pathological entities and, especially, on the microscopic features of the OA, which clearly show an intimate relationship between the ameloblastoma and the mineralized tissues.

The histopathological features of the OA are complex. There is a proliferating odontogenic epithelium portion similar to that of an ameloblastoma, generally presenting a plexiform or follicular pattern. This epithelial portion appears intermingled with dental tissues of variable degrees of maturity in the form of developing rudimentary teeth, resembling a compound odontoma or conglomerate masses of enamel, dentin and cementum, as seen in a complex odontoma.[1] The cases presented here exhibited all the histopathological features of OAs. In both cases, the dental tissue was represented by masses of dentin and cementum. Enamel matrix, however, was not observed.

From a clinical and radiographic point of view, differential diagnoses include several odontogenic and non-odontogenic lesions exhibiting well-defined uni- or multilocular radiolucencies with varying amounts of radiopaque material within them. These include developing compound or complex odontomas, ameloblastic fibroodontomas, calcifying epithelial odontogenic tumors, calcifying odontogenic cystic tumors, adenomatoid odontogenic tumors and cementoossifying fibromas. Based on the clinical and radiographic findings of the first case presented here, the differential diagnosis hypotheses included an adenomatoid odontogenic tumor, a calcifying epithelial odontogenic tumor and an ameloblastic fibroodontoma. However, the resorption of adjacent tooth roots observed in this case is not usually observed in cases of an adenomatoid odontogenic tumor. Owing to a possible diagnosis of a calcifying epithelial odontogenic tumor, which

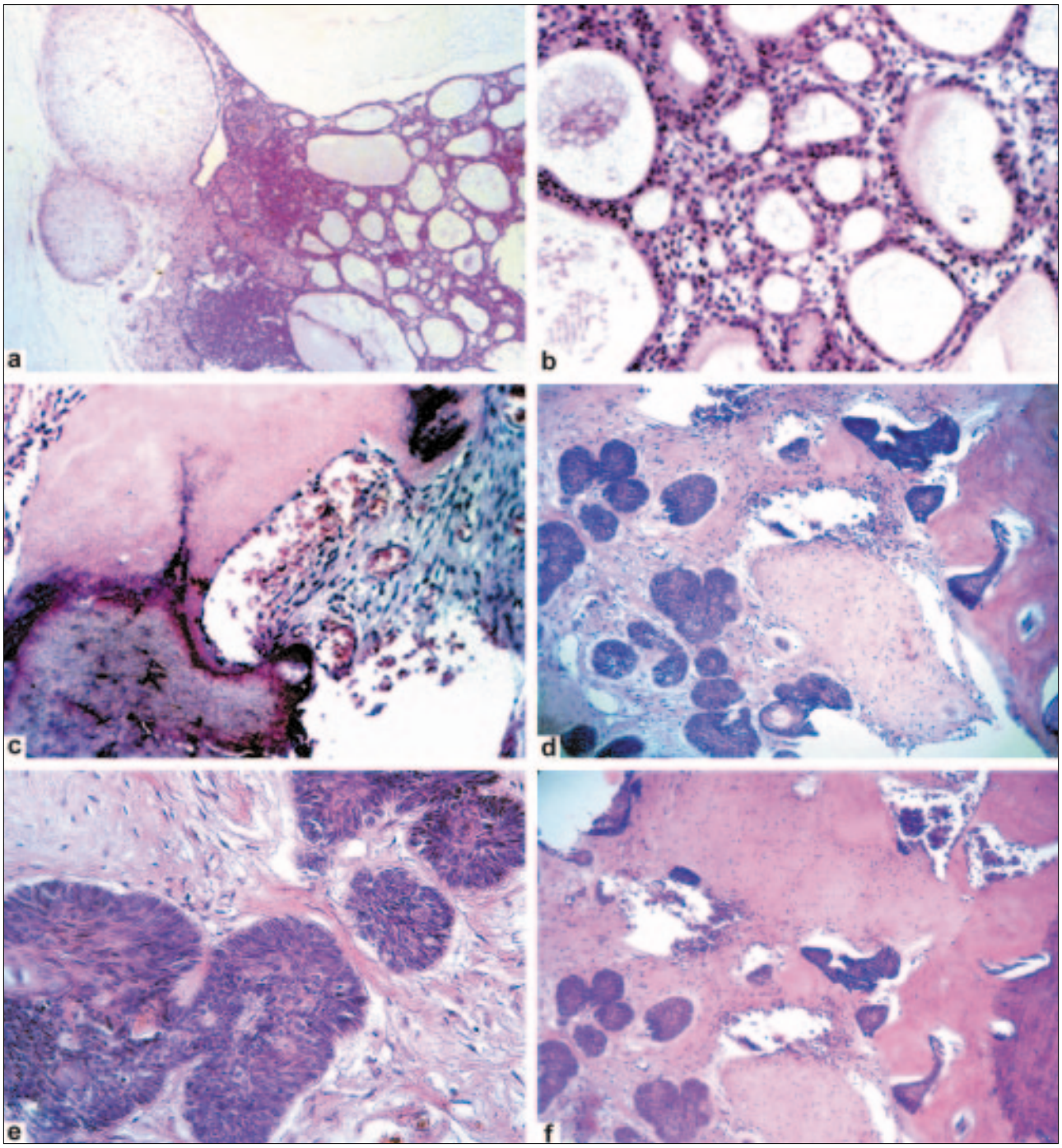

Figure 3: Histopathological examination. (a–c) First case; (d–f) Second case. (a) Odontogenic epithelium. (b) Proliferating odontogenic epithelium. (c) Calcified structures: Dentine (eosinophilic structure) and cementum (basophilic structure) of variable degrees of maturity. (d) Sheets of odontogenic epithelium. (e) Proliferating odontogenic epithelium. (f) Calcified structures in the mesenchymal component: Immature dentine (eosinophilic structure) and cementum (basophilic structure). H and E, original magnification, ×100 (a, d, f) and ×200 (b, c, e)

behaves as an ameloblastoma, the tumor was surgically enucleated with a 2 mm safety margin.

Although located in the posterior mandible, the second case presented in this study resembled that of an odontoma and, for this reason, it was only enucleated. No recurrences of the tumor were observed after a follow-up period of 6 years in the first case and 6 months in the second case.

In the review by Mosqueda-Taylor *et al.*,[7] 3 of 14 cases recurred (21.4%). These authors emphasized that OA should be closely followed-up for at least 5 years. The patient of our second case should therefore be closely followed-up for a yet longer period of time.

The potential for OA to recur is well known. In fact, Yamamoto *et al.*[13] demonstrated a high proliferation

potential of the OA based on the expression of tenascin in the basement membrane of the odontogenic epithelium of this tumor and on the results obtained with proliferating cell nuclear antigen (PCNA) staining. These results indicate that this tumor may have the same biologic potential as that of an ameloblastoma and should therefore be treated and followed-up in a similar fashion.[1,7]

Because of the rarity of OAs and its similarity to other odontogenic lesions, a pre-operative diagnosis is difficult to achieve based only on the clinical and radiographic features of the lesion.[7] Nonetheless, surgeons should be aware of the existence of these odontogenic tumors in order to properly treat and follow-up patients who might present them.

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