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Case Report

Odontoameloblastoma of Maxilla - A Rare Odontogenic Entity Mimicking Fibro-osseous Lesion: A Case Report

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Abstract

Odontoameloblastoma (OA) is an exceedingly uncommon odontogenic tumor that contains an ameloblastomatous component and odontoma like elements. It is characterized by slow-growing lesion with growth pattern similar to solid multicystic ameloblastoma. The majority of the tumors are associated with unerupted teeth and commonly seen in males. It is usually asymptomatic and may occur in either maxilla or mandible but shows a slight predilection for the mandible. As this tumor is extremely rare, there exists controversy regarding its development and treatment. Here, we present a case of OA in 20-year-old male patient resembling a fibro-osseous lesion and a brief review of the related literature.

Key words: Maxilla, Odontoameloblastoma, Odontogenic tumor, Odontoma

INTRODUCTION

The odontoameloblastoma (OA) is an extremely rare neoplasm, which is defined in the current WHO histological classification of odontogenic tumors as a tumor that "includes odontogenic ectomesenchyme in addition to odontogenic epithelium that resembles an ameloblastoma both in structure and behavior."[1]

It usually occurs between 6 months and 40 years predominantly involving mandible with a predilection for molar-premolar region.[2,3] It is an expansile centrally destructive lesion exhibiting slow growing characteristics like ameloblastoma and if left untreated may cause substantial facial deformity. Symptoms include a slowly progressive swelling of the alveolar plates, dull pain, an altered occlusion, delayed eruption, or impacted teeth. Radiologically the tumor presents as a well-defined unilocular or multilocular radiolucent cavity containing varying amounts of radiopaque material, which may or may not bear resemblance to formed teeth. It may also be in the form of small, dense particles or as a large central mass leading to divergence of adjacent tooth roots.[2,4]

We are presenting a case of OA in the posterior region of the right maxilla of a 20-year-old male.

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CASE REPORT

A 20-year-old male reported with a chief complaint of a slow growing swelling in his right mid face region for 1 year (Figure 1a). The swelling was asymptomatic but was causing discomfort while speaking and eating. The swelling was gradually increased in size and reached to present size. Intraoral examination revealed a diffuse swelling on palate extending from 11 tooth region to the right maxillary tuberosity obliterating the vestibule. There was the absence of 13, 15, 16, 17 teeth for last 6 months. The swelling was firm, nontender, nonfluctuant, nonpulsatile, and smooth on palpation. The overlying mucosa overlying mucosa was normal in color and texture.

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Computed tomography scan showed a diffuse lesion that produced marked destruction of bone and contained abundant dense and irregular foci of mineralized tissue surrounded by radiolucent areas. The tumor extended upwards to involve the floor of the orbit. Displacement of the lateral nasal wall with obliteration of maxillary sinus (Figure 1b).

The patient was advised for biopsy, and the histopathological feature revealed multiple ameloblastic islands arranged in follicles in a scanty fibro-connective tissue stroma containing numerous small and large areas of calcified mass resembling dental hard tissues such as dentine, osteodentin, cementum with a variable amount of primitive mesenchymal tissue (Figure 2). A histological diagnosis of

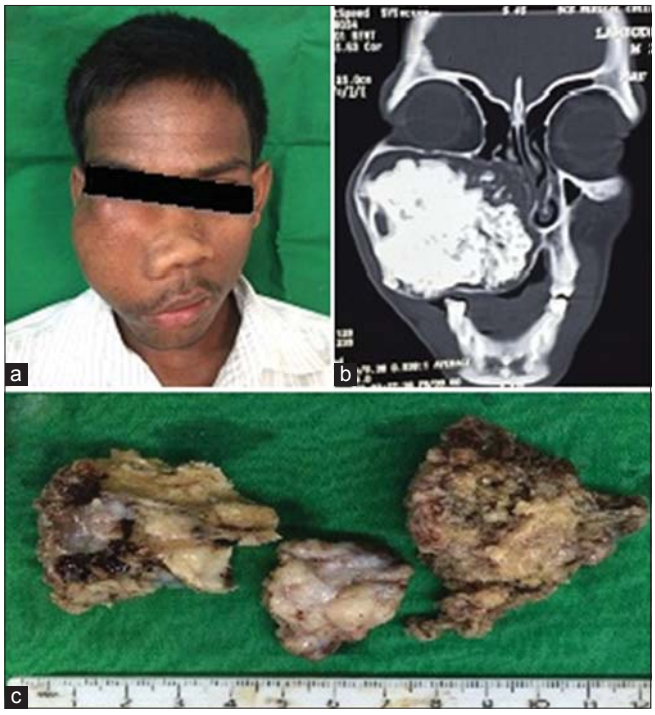

Figure 1: (a) Clinical picture. (b) Computed tomography scan axial view showing extension of the lesion. (c) Excised gross specimen

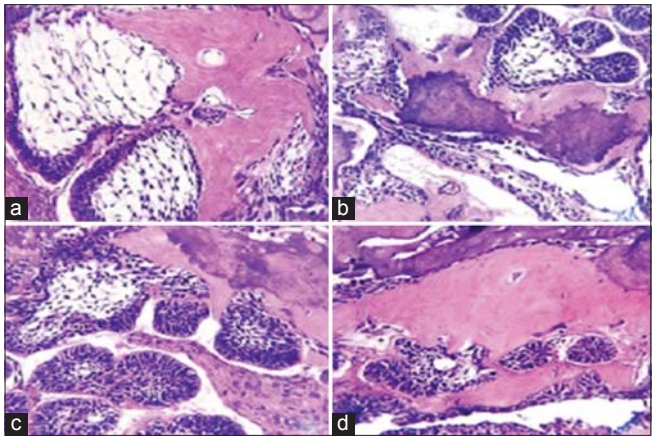

Figure 2: (a-d)-Histopathology (H and E, ×400)

OA was given. Then, the patient was operated at a later date, and the tumor was excised. The gross specimen (Figure 1c) was showing multiple grayish white hard and soft tissue specimen (m) about 6 cm × 3 cm. On microscopic examination of excised tumor showed the features of OA same as in incisional biopsy specimen. Hence, the final diagnosis confirmed as OA of right maxilla. The post-operative recovery was uneventful, and after 1 year follow-up, the patient was normal with no evidence of recurrence.

DISCUSSION

The OA, also known by some authors as ameloblastic odontoma,[5,6] 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.[1,4] The epithelial proliferation forms ameloblastic islands in follicular or plexiform patterns typical of ameloblastoma but, unlike conventional ameloblastoma, these induce the production of mineralized dental tissues on the adjacent mesenchymal cells and may respond to this changes with the production of enamel.

Although there are various case reports of this condition, several authors agree that only a few of them met the histologic and clinical criteria to be classified as OA,[4,5,7,8] which is supported by the present study. 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.[9] Kaugars and Zussmann[5] have suggested following criteria for the histological diagnosis of OA - (a) unequivocal ameloblastoma, (b) connective tissue with a mature homogenous appearance, and (c) fragments of malformed calcified dental structures.

The case presented here exhibited all the histopathological features of OAs. In this case, 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, the differential diagnosis includes several odontogenic and non-odontogenic well-defined unilocular or multilocular radiolucencies with varying amounts of radiopaque material within them. These include developing compound or complex odontomas, ameloblastic fibro-odontoma, calcifying epithelial odontogenic tumor, calcifying odontogenic cyst, adenomatoid odontogenic tumor, and cemento-ossifying fibroma. OA radiologically mimicking

Table 1: Clinico-pathological comparison between odontoameloblastoma and ossifying fibroma

Odontoameloblastoma	Ossifying fibroma
1. Age - 6 month-40 years	1. 30-40 years
2. Sex - Male>Female	2. Female>Male
3. Site - Mandible>maxilla	3. Mandible>maxillaoma
4. C/F-painless mass associated with a delayed eruption	4. Asymptomatic expansile slow growing
5. Biological behavior-locally aggressive	5. Benign behavior
6. Radiographic-well-defined unilocular/multilocular radiolucency having varying amounts of radiopaque material resembling dental hard structures with the features of a complex odontoma	6. Well-defined circumscribed border, expanded cortex, heterogeneous calcification medullary pattern
7. Histopathology-ameloblastoma with odontoma like features	7. Cellular fibrous stroma, islands of new bone formation, relatively homogenous pattern
8. Treatment-enucleation and resection	8.Curettage/excision
9. Prognosis-high rate of recurrence	9. Recurrence – rare
10. Source-odntogenic origin	10. Non-odontogenic

with fibro-osseous lesion like ossifying fibroma, but here, we try to speak out some clinico-radiological feature to differentiate OA from ossifying fibroma (Table 1).

The pathogenesis of OA is unknown. One possible explanation is that the mineralized dental tissues are formed as a hamartomatous proliferation in response to inductive stimuli produced by the proliferating epithelium over the mesenchymal tissue.[4]

Wächter *et al.*[10] compared four cases of OA with 14 cases of ameloblastic fibro-odontoma and found that there are no clear cut histological criteria to separate these two lesions. However, solid/multicystic ameloblastoma like structures was more characteristic for the OA, whereas ectomesenchymal component was more pronounced in the ameloblastic fibro-odontoma.

The potential for OA to recur is well known. In fact, Yamamoto *et al.*[11] 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 staining 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.[9,12] In the review by Mosqueda-Taylor *et al.*,[12] 3 of 14 cases recurred (21.4%), which was a similar figure to that found by Reichart *et al.*[13] for ameloblastoma.

CONCLUSION

OA may be clearly recognized and distinguished from other mixed odontogenic tumors. Although it tends to occur at a prior age than conventional ameloblastoma, it has the same potential to produce bone expansion, root resorption, and recurrence. Pre-operative clinico-radiological diagnosis is more challenging than histopathological diagnosis for suitable treatment planning. OA should be treated as locally aggressive lesions with periodic follow-up.

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