10.5005/jp-journals-10024-1255

CASE REPORT

Adenoameloblastoma: A Dilemma in Diagnosis

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ABSTRACT

Adenoameloblastoma or adenomatoid odontogenic tumor (AOT) is an uncommon, benign, epithelial lesion of odontogenic origin. It is a rare benign odontogenic tumor of the jaw affecting mostly young individuals with predominance in female. It occurs mostly in maxillary anterior region. On the basis of clinical and radiographical picture, it is often misdiagnosed as an odontogenic cyst.

We report on a rare case of a 13-year-old male patient with a follicular variety of AOT in mandibular left anterior region which is unusual for the same. Clinically and radiographically, the lesion was mimicking as a dentigerous cyst. Later surgical enucleation was done and specimen was sent for microscopic examination and was diagnosed as AOT along with a dentinoid-like deposits which is a rare finding.

Keywords: Adenoameloblastoma, Dentinoid, Adenomatiod odontogenic tumor, Impacted tooth, Mandible.

How to cite this article: Laheji A, Sakharde S, Chidambaram S, Gondhalekar RR, Shankar U, Radhika A. Adenoameloblastoma: A Dilemma in Diagnosis. J Contemp Dent Pract 2012;13(6): 925-929.

Source of support: Nil

Conflict of interest: None declared

INTRODUCTION

The adenoameloblastoma is a nonaggressive, benign, hamartomatous, slow but progressively growing lesion of odontogenic origin.[1] It was first described by Dreibaldt in 1907 as a pseudoadenoameloblastoma.[2] Harbitz in 1915 reported it as adamantoma and Bernier and Ticke were first to publish a case using the name adenoameloblastoma.[3] The term adenomatoid odontogenic tumor (AOT), which is being widely used, was given by Philipsen and Birn in 1969 and was opted by World Health Organization (WHO) classification of odontogenic tumor in 1971.[4]

The three clinicopathological variant of AOT are follicular type (73%), extrafollicular type (24%) and peripheral variety (3%), all with identical histology.[5] The follicular type is a central intraosseus lesion associated with an impacted tooth. The extrafollicular intraosseus AOT often located between, above or superimposed upon the roots of adjacent erupted teeth and has no relation with an unerupted tooth. The peripheral variant appears as a gingival fibroma or epulis attached to the labial gingiva.

In the latest edition of WHO classification of odontogenic tumors in 2005, AOT was classified into the first group of tumors (odontogenic epithelium without ectomesenchyme) instead of the second group (odontogenic epithelium with ectomesenchyme).[6] Because of the absence of ectomesenchyme in immunohistochemical staining, dysplastic dentin in AOT is now considered as the result of a metaplastic process rather than epithelial-ectomesenchyme interaction.[6]

The reported case is of a 13-year-old male patient with follicular AOT in mandibular anterior region which was mimicking clinically and radiographically as a dentigerous cyst, but histopathological features resemble AOT with hard tissue formation within it.

CASE REPORT

A 13-year-old male patient reported to the department of oral medicine and radiology with complaint of swelling in lower left anterior region of the jaw since 6 months. The swelling was painless and increasing gradually in size. The patient was apparently healthy, with no significant history related to the lesion and was not taking any medication for it.

Extraoral examination revealed facial asymmetry with a single, oval, well-demarcated, nontender, bony hard swelling in the mandibular left anterior region (Fig. 1). The swelling was extending from corner of mouth to inferior border of mandible superoinferiorly and 1 cm lateral to chin to half the body of mandible. The swelling was around 4 × 3 cm in size. There was no sign of numbness over anterior region of jaw.

Intraoral examination revealed a firm swelling over mandibular left anterior region extending from 32 to 35 regions, obliterating labial and buccal vestibule with a normal overlying mucosa (Fig. 2). A total of 74 was retained and was vital and immobile and 34 was unerupted.

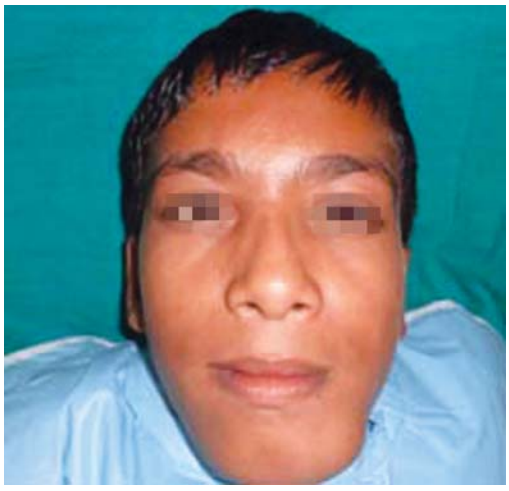

Fig. 1: Clinical photograph showing extraoral swelling in mandibular left anterior region

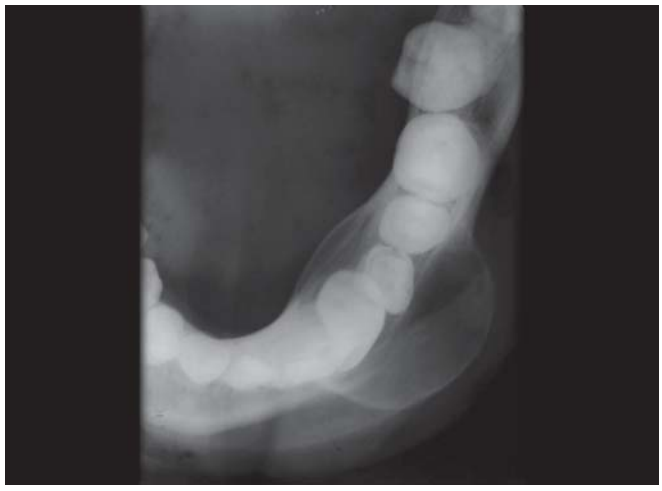

Fig. 3: Lateral mandibular occlusal radiograph revealed buccal and lingual expansion of cortical bone

Occlusal radiograph demonstrated that the lesion was considerably expansile with labiolingual expansion and there was the thinning of the labial and lingual cortical plates (Fig. 3).

Panoramic radiograph demonstrated a unilocular, round, radiolucent lesion of size 3 × 3 cm, enveloping the impacted 34 is seen in left mandibular anterior region. The lesion was surrounded by well-defined radiopaque border with sclerosis in some area. The lesion was extending up to the lower border of mandible in canine and premolar region leading to thinning of outer cortex. There was no root resorption, but the displacement of 31, 32, 33 was present (Fig. 4).

The provisional diagnoses made for the lesion was dentigerous cyst and differential diagnosis made were unicystic ameloblastoma and AOT.

On aspiration with 18-gauge needle, blood-mixed fluid was obtained and was sent for histological examination and it was reported as an inflammatory cyst.

Blood examination revealed normal findings. The patient was operated under local anesthesia. A total of 74

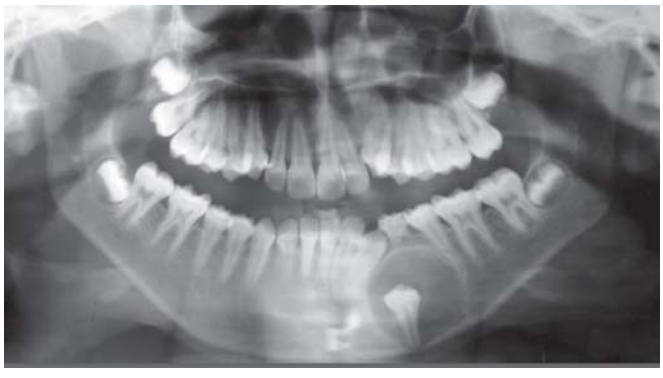

Fig. 4: Panoramic radiograph revealed a well-defined radiolucent lesion along with impacted 34

was extracted followed by a full thickness periosteal flap was raised from mandibular left 31 to 36 in the buccal vestibule. The buccal cortex was resorbed with a thin shell of bone in between. The lining of the cystic lesion was carefully separated from the mucoperiosteum and the lesion was enucleated along with unerupted first premolar. The tumor cavity was irrigated with saline and betadine and later was filled with gel foam and wound was sutured with 3-0 vicryl.

The healing was uneventful and the patient is under observation since 3 months.

The excised mass was brownish in color and measuring about 3 × 2 × 2 cm in size. There was thick brownish fluid present within the mass. The histopathologic (H-E stain) report revealed a cystic cavity lined by stratified squamous epithelium with multinodular proliferation of spindle, columnar and cuboidal cells arranged in the form of whorled mass (Fig. 5). Few calcifications were also evident. The tumor also exhibited large amounts of eosinophilic matrix material in the stroma, consistent with dentinoid. An irregular calcified mass of dentin with clear dentinal tubules was seen in one of the sections (Fig. 6). Formation of enamel matrix and primitive appearing connective tissue was not present. Periodic acid-Schiff staining was positive for it

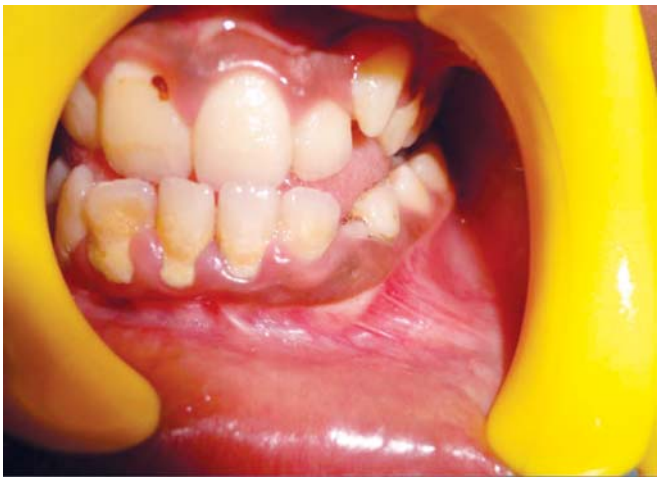

Fig. 2: Intraoral photograph showing obliteration of labial vestibule

(Fig. 7). Congo red staining was positive for amyloid (Fig. 8). Histopathological diagnosis of follicular AOT with dentinoid formation was made.

DISCUSSION

AOT is a rare benign odontogenic tumor of epithelial origin consisting of 2.2 to 7.1%[3,7,8] of all odontogenic tumors and 0.1%[9] of tumors and cysts of jaw. Follicular and extrafollicular variants together are more commonly found in the maxilla than in the mandible (2.9:1 ratio).[8] More than two-thirds are diagnosed in the second decade, mostly in the 13 to 19 years age group.[10-12] The female: male ratio is 2.4:1.[12] The tumor is usually associated with unerupted teeth, frequently canines or lateral incisors.[3,7,13] The relative recurrence of AOT is very low.[13]

Here, the reported case is of a 13-year-old male patient with AOT associated with an unerupted mandibular premolar which is very rare (0.16%).[12] The tumor is slow growing, painless and may expand the cortical plates but invasion of soft tissue does not occur.[12]

Radiographically the follicular variety is characterized as a well-defined, unilocular radiolucent lesion surrounding the crown and often part of the root of an unerupted tooth. Borders of the lesion are corticated and sclerosis may be observed.[14,15] Khot and Vibhakar[2] and Haider[16] also reported case with similar radiographic appearance.

Radiographically, radiopacities develop in about two thirds of cases.[14] Intraoral radiographs may be required to demonstrate the calcifications within the lesion, which may not be seen on panoramic radiographs.[13,14] In the reported case, in occlusal and panoramic radiographs, there were no radiopacities seen but periapical radiograph of the excised mass revealed significant radiopacities within the lesion. Garg et al[3] reported a case in which there are foci of radiopaque mass seen within the radiolucent lesion in radiograph. But there are cases where the lesion has no

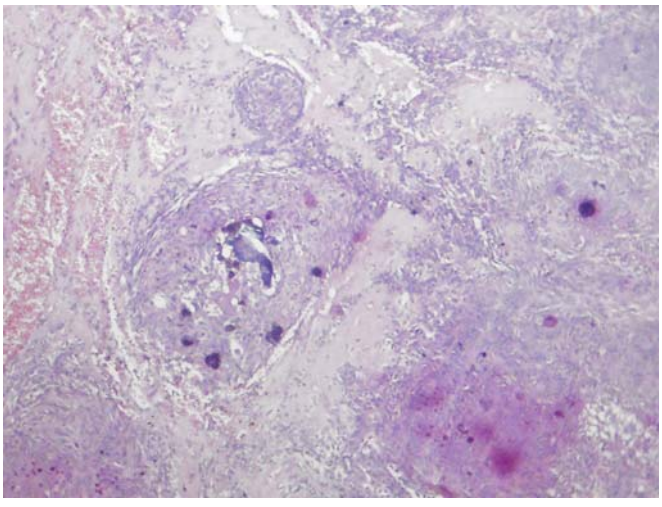

Fig. 5: Microscopic image (hematoxylin-eosin, original magnification ×10) of adenomatoid odontogenic tumor, revealing nodules of basophilic cells with adenomatoid structures

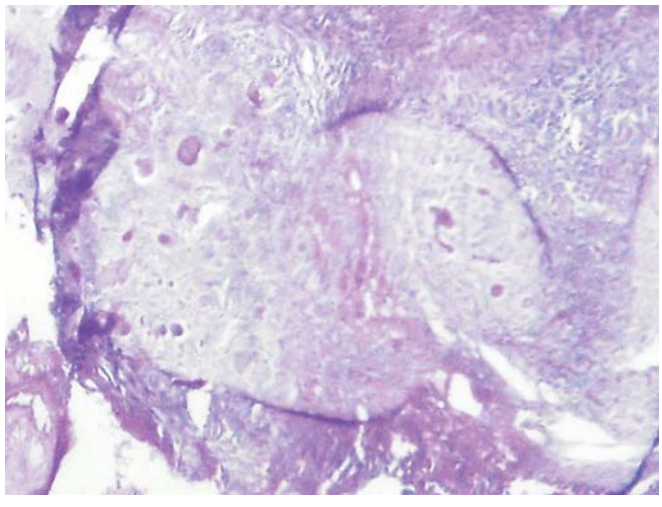

Fig. 7: Microscopic image (periodic acid-Schiff, original magnification ×10) of adenomatoid odontogenic tumor, revealing nodules of adenomatoid structures

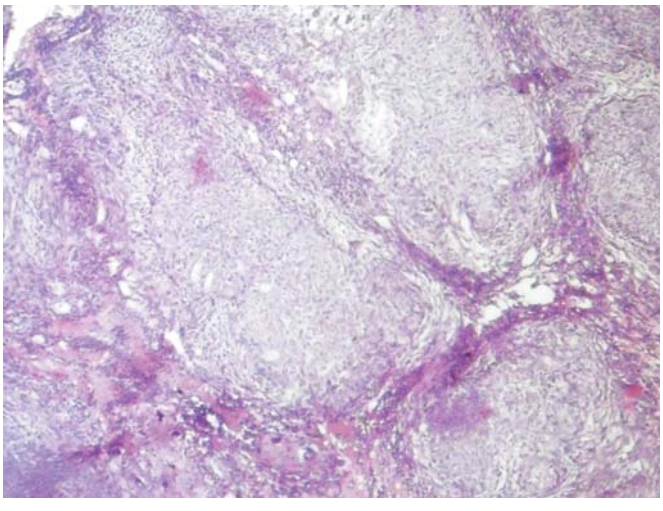

Fig. 6: Microscopic image (hematoxylin-eosin, original magnification ×40) of adenomatoid odontogenic tumor with dentinoid-like material

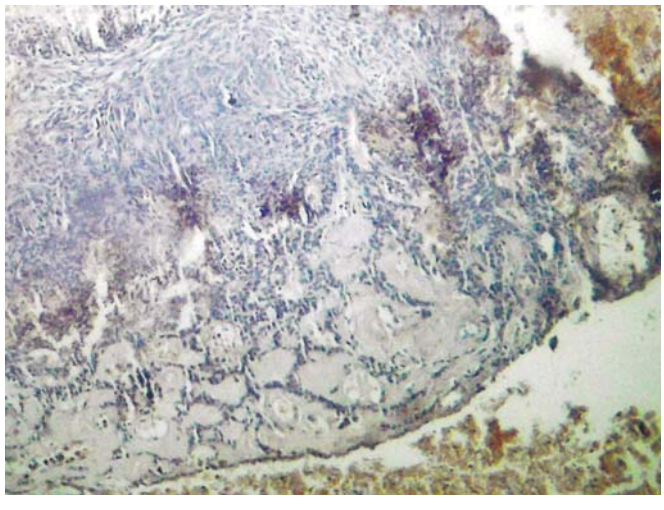

Fig. 8: Microscopic image (congo red, original magnification) of adenomatoid odontogenic tumor revealing amyloid in lesion

radiopaque component in such cases, a dentigerous cyst is a preferred differential diagnosis.[2,7]

Root resorption is usually not seen in AOT, rather the displacement of adjacent teeth is seen which was evident in our case also. Though some cases of root resorption is also evident.[2,3,17]

In the reported case blood-mixed aspiratory fluid was obtained. Nigam et al[17] also reported blood as aspiratory fluid, whereas Panjwani et al[15] reported straw-colored fluid on aspiration. Histologically, the most striking pattern is that of various sizes of solid nodules of columnar or cuboidal epithelial cells forming nests or rosette-like structures with minimal stromal connective tissue. There may be nodules of polyhedral, eosinophilic epithelial cells with squamous appearance and exhibiting well-defined cell boundaries and prominent intracellular bridges. These islands may contain pools of amorphous amyloid-like material and globular masses of calcified material suggesting it as a hybrid tumor.[18,19] In the adenomatoid odontogenic tumor, hard tissues may occur as dysplastic dentin, enamel matrix and dystrophic calcifications.[1] In the reported case, there was dystrophic calcification along with dentinoid formation was observed.

WHO has classified it under benign odontogenic tumor of odontogenic epithelial origin without hard tissue formation, which is contradictory for the reported case.[6]

The most common non-neoplastic lesions of jaw in this age group are apical cyst, dentigerous cyst, calcifying epithelial odontogenic cyst, odontogenic keratocyst, periapical granuloma and central giant cell granuloma. The radiographic findings of AOT frequently mimics other odontogenic lesions, such as dentigerous cysts, calcifying odontogenic cysts, calcifying odontogenic tumors, globulomaxillary cysts, ameloblastomas, odontogenic keratocysts and periapical disease.[2-5,7,9,13-17] Thus, it makes dilemma in diagnosing a lesion.

CONCLUSION

As AOT is a rare tumor of second decade of life; therefore, it's been mostly overlooked while making a diagnosis. Its clinical and radiographical features may bring dilemma for diagnosis as it mimics other lesion like dentigerous cyst. The final diagnosis of the AOT was made after histological examination of the excised mass. Hence, we should be careful while making diagnosis of such similar type of lesion.

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