An Extrafollicular Cystic Adenomatoid Odontogenic Tumor of the Mandible Associated with Clear Cell Calcifying Epithelial Odontogenic Tumor: A Rare Case Report

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ABSTRACT

Background: The adenomatoid odontogenic tumor is a relatively rare benign epithelial odontogenic tumor. It contains both epithelial and mesenchymal components. Few cases presented as an extrafollicular lesion or involve the mandible or associated with other odontogenic lesions. This paper represents a rare case of an extrafollicular AOT.

Case presentation: A 24-year-old female had a painless swelling on the right side of the lower jaw since one-month duration. Intraorally there was a well defined fluctuant-blue swelling in the right alveolar premolar region measuring 1×2 cm obliterating the right lower buccal vestibule. Grade II mobility in the vital 44 and 45 teeth were observed. Panoramic radiographs showed a well-defined pear shaped radiolucent lesion without calcifications between the roots of 44 and 45 that cause roots divergence. The lesion totally enucleated with the tooth 44 which showed root resorption. Microscopically, a cystic lesion lined by simple odontogenic epithelial cells with areas of polygonal nodular proliferation (sheets, whorled masses with few duct-like structures and clear cells) and amorphous eosinophilic material. This material was PAS –negative, and Congo red positive. The case diagnosed as a cystic extrafollicular variant of AOT with CEOT-like areas.

Conclusions This case report describes the first known case of extrafollicular combined AOT/CEOT associated with root resorption and clear cells. The existence of clear cells suggest a possible aggressive nature and long follow up recommended.

Keywords: Adenomatoid odontogenic tumor, calcifying epithelial odontogenic tumor, clear cells, mandible. (J Bagh Coll Dentistry 2016; 28(2):47-51).

INTRODUCTION

Adenomatoid odontogenic tumor (AOT) is a relatively rare epithelial odontogenic tumor [(1)]. It is regarded either as a true benign, non-aggressive non-invasive tumor or as a developmental hamartomatous odontogenic growth. It is believed that the lesion is not a neoplasm. The World Health Organization (WHO) defined AOT as "A tumor of odontogenic epithelium with duct-like structures and with varying degrees of inductive changes in the connective tissue. The tumor may be partly cystic, or contain solid masses in the wall of a large cyst [(1)]. Its ICD-O code is 9300/0. According to the biologic classification, AOT is a benign tumor with no recurrence potential [(2)].

AOT is more frequently reported in females, and less commonly involve the mandible [(3,4)]. A percentage of 26.9 of AOT cases may present as an extrafollicular lesion with M: F ratio of (close to) 1:2 [(3)]. It may locate between, above or superimpose roots of erupted teeth [(5)]. Histologically, AOT has diverse features [(6,7)].

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The literature review remarked for the association of AOT cases with another odontogenic lesion [(8,9)].

Several investigators have pointed out the presence of CEOT-like tissue in AOT [(10-13)] and believed to be a frequent finding [(14)].

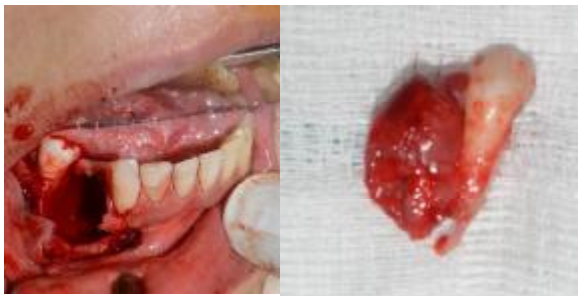
There is still a need to continue reporting well-documented cases of the extrafollicular variant. Concerning the exact location especially when they seem to have unusual or rare' histomorphological features, or in a new ethnic group or geographical region and relate these findings to the biological behavior of AOTs and possibility of tumor recurrence.

CASE REPORT

A 24-year-old female attended the Maxillofacial Center in the teaching hospital in Sulaimani on 22/4/2013 with a chief complaint of a painless swelling on the right side of the lower jaw since one-month duration. The patient was clinically healthy, and all her vital signs were within normal limits. On clinical examination, there was a well-localized swelling in the right alveolar premolar region measuring 1×2 cm obliterating the right lower buccal vestibule. Grade II mobility in 44 and 45 were observed. The swelling was fluctuant, and the overlying

mucosa was blue in color. Neither the teeth nor the swelling was tender, and there was no palpable lymph node in the submandibular region. Electric pulp test indicated vital teeth. The patient Involve the right side alveolus between the roots of 44 and 45and cause roots divergence. The root of 44 showed resorption (Figure-1C). FNA revealed a thin clear fluid.

Excisional biopsy of the mass along with the tooth 44 was done (figure 1B). There were no apparent infiltrations of the surrounding bone. The surgical sample sent for histopathological examination. Grossly, the specimen consisted of a cystic lesion attached to the lateral surface of the tooth 44, with brownish wall and it contained thin clear fluid. The specimen measured approximately 1 x0.5 x2 cm in dimension. Two general histopathologists from two centers assessed the slides. The results were dissimilar. In the first report, the case was diagnosed as CEOT while the second indicated mucoepidermoid carcinoma. Therefore, a third opinion was suggested to be done by an oral pathologist.



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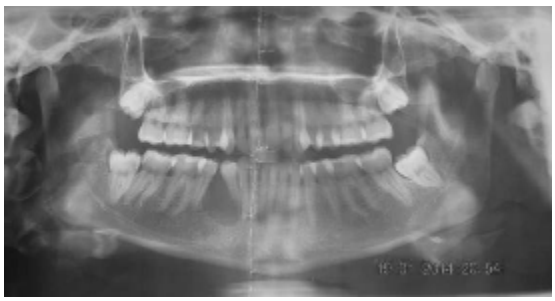

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Figure1: Extirpated tumor completely encased the tooth (A). The cyst bag adhered laterally to the lower first premolar's root (B). Panoramic radiograph before therapy. A well-demarcated unicystic radiolucent lesion in the lower right side jaw between the roots of the teeth 44 and 45 results in root divergence, and adjacent tooth 44 showed resorption of the root (C).

Histopathologically, the cyst lined by thin odontogenic epithelium cells that surround a loose fibrovascular stroma. The epithelial cells extend shortly as strands intramurally or show thinking with hyalinization or continued with lace-like and nodular proliferation (figure 2). The proliferating cells revealed: few duct-like structures, polyhedral cells that had a clear or sometimes intensely eosinophilic cytoplasm, with nuclear hyperchromatism and a mild degree of variability in nuclear size (pleomorphism) in a scant connective tissue stroma. In the intercellular and intraductal sites, there are amorphous eosinophilic materials. The cystic wall is thick connective tissue capsule exhibiting bundles of loosely arranged collagen bundles with fibroblasts, blood vessels and areas of hemorrhage. Therefore, the case suggested containing clear cell variant of CEOT. Then the slides subjected for Congo red and PAS stains to identify amyloid and confirm the diagnosis. The material stained positively with Congo red and negatively with PAS. The final diagnosis was a cystic variant of extrafollicular types AOT with CEOT-like areas

The patient was followed-up for two year and six months without any evidence of recurrence (figure -3). Then inform consent was obtained from the patients to publish the work. The paper was approved by the ethics committee in the Medical Faculty.

DISCUSSION

The present report recorded the first case of extrafollicular combined AOT/CEOT in an Iraqi-Kurdish female patient. The case showed distinctive clinical and histological features.

Comparing the clinical and radiological features of our case with AOT and combined AOT/CEOT cases described in the literature indicate that the sex and the age of the patient are similar. The patient's age fit with the peak incidence of AOT tumor (the second decade of life). Nevertheless, patients with extrafollicular AOT have been reported to be significantly in older age than those with the follicular variant [(3)].

Philipsen and coauthors [(4)] conducted a collaborative retrospective study that included 1082 AOT cases from 12 major centers in the world. They collected the data up to the end of 2005 and showed that 26.7% (9% male, 17.9% female) of AOT were the extrafollicular type with slight maxillary predominance 15.5% in comparison to mandible 11.2% [(4)]. On the other hand, Becker et al. [(3)] also find that extrafollicular variant AOT constituted 27%. However, they

reported opposing localization. They found (12%) of cases in the maxilla and (15%) of cases in the mandible. Concerning the reported combined AOT/CEOT cases, Siar and Ng indicated that such cases had a predilection for the mandible (55.6%) [15].The literature indicated that the reported combined AOT/CEOT lesions were predominantly follicular [11,13,16].

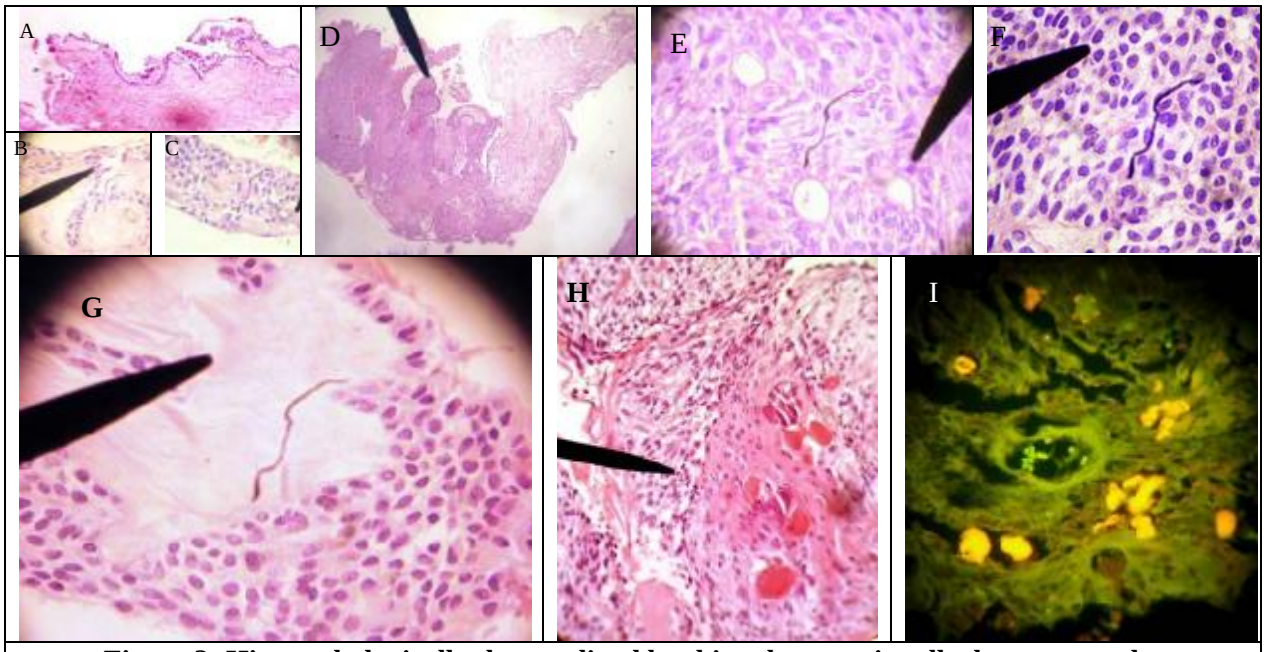

Figure 2: Histopathologically the cyst lined by thin odontogenic cells that surround a fibrovascular stroma (A). The epithelial cells extend as strands intramurally or show thinking with hyalinization (B &C), beside an area of nodular proliferation (D arrow). The proliferating cells revealed: few duct-like structures (E), polygonal clear cells with nuclear hyperchromatism (F) and polymorphism (G), and foci intercellular and intraductal eosinophilic material (H). The amorphous material is amyloid. After staining withCongo red it appears bright orange under fluorescent light (I).(A and D, X10; remaining photomicrographs X40)

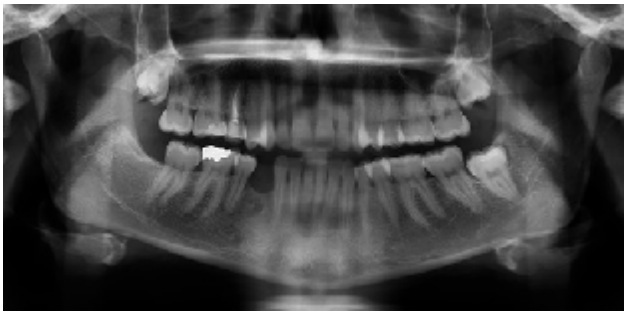

Figure 3: Panoramic radiograph of the patient 54 months after treatments

In contrast, the extrafollicular AOT cases showed histologically classic AOT feature [17-19] and involved the lower jaw. The present case and that of Wilia [18] are in the canine-premolar region while Yilmaz case [19] located in the mandibular anterior region. However, the current case differ from them in type. It is an extrafollicular combined AOT/CEOT lesion.

AOT is a slowly growing tumor and when detectable at the small size described as a localized swelling of the involved jaw. It has been indicated that the size of an AOT influenced by the patient's age [3]. Our patient was 24 years old, her lesion is 1x2 cm and become evident within one month only. The unexpected short disease duration is unlike the documented clinical registered period in the literature which was mostly between 6-12months disease duration, even in young aged patients [9,10,17].

According to the radiographical findings, extrafollicular AOT has several topographical relation to the teeth. Herein, the case is categorized as E2 type; it indicates an inter-radicular localized tumor with extensive growth

causing divergence of roots [5]. However, our case associated with root resorption. Irregular root resorption is rarely seen with intraosseous AOT. Identification of radiological root resorption was related to a longer disease duration and to be associated with patients aged 30 years and above [3]. As well as it also reported with unusual large follicular lesions (more than 3 cm) [6]. However, in one of these large-size reported cases, the lesion identified within six months in 19 years old male [6]. Furthermore, Garg et al. linked evidence of root resorption with fast-growing, unencapsulated AOT lesion [20]. The above explanations can not be applied to our case since it had short disease duration, small size and histologically well capsulated.

The gross findings of our case were consistent with earlier descriptions of extrafollicular AOT in the literature. Microscopically, AOT can form a hybrid growth with a histo-architectural variety containing both epithelial and mesenchymal components [6,7] or even associated with or arising from other odontogenic lesions especially in a young patient [9]. There are several reports mentioned a combined AOT/CEOT lesions [11-13,16]. Mosqueda-Taylor et al. believed that such findings have no clinical or prognostic differences on the common AOT. They considered the occurrence of areas of CEOT-like tissue in "classic" AOT is a spectrum of AOT since there is no typical pleomorphism of the true CEOT and the polyhedral cells confined to the nodular areas near the cyst lining and tend to produce mineralized tissue [14]. They stated that AOT cases without CEOT-like areas were those who had few calcified material. However, our case contained few cells with pleomorphism and areas of clear cells within the CEOT-like cells, which is an uncommon finding. Beside that radiographically there were no calcified foci. Tiny calcification is better to be examined by intraoral periapical radiographs.

It is confirmed clinically and histologically that AOT is a hamartoma. The existence of few tumor-like nodules (epithelial proliferation) that have inductive capacity in the cystic lesion is no more regarded as neoplasm growth despite their slow-growing property [2,6]. Nevertheless, it is worth to mention that the existence of clear cell CEOT variant within the growth may suggest aggressiveness and associated with both root and cortical perforation and fast clinical swelling as seen in this case.

AOTs alone or with CEOT is a benign, encapsulated lesion, and conservative surgical enucleation or curettage is the treatment of choice. Consequently, the tumor may be conservatively removed, with little or no danger of recurrence. For large tumors, subtotal resection may be necessary [2,21].

As conclusion; this case report describes the first known case of extrafollicular AOT associated with root resorption and containing clear-cell CEOT-like area. Since CEOT with clear cell changes is known to recur more frequently, a life-long follow-up of such patient may be required.

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