Extrafollicular Adenomatoid Odontogenic Tumour

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Abstract

Adenomatoid Odontogenic Tumour (AOT) is an unusual benign tumour unique to maxillofacial area, with a tendency to involve the tooth bearing areas of both the jaws and accounting for 3% of all the odontogenic tumours. Diagnosis of the tumour is sometimes intriguing owing to its variable clinical and radiographic presentation. One such variable and relatively rare presentation of extrafollicular type of adenomatoid odontogenic tumour in anterior maxilla of an eighteen year old female has been discussed in this paper which can be refreshing for the practitioner.

Keywords : Adenomatoid Odontogenic Tumour, Extrafollicular type

INTRODUCTION

Adenomatoid odontogenic tumour (AOT) is a relatively uncommon distinct odontogenic neoplasm that was first described by Steensland[1,2] in 1905 and by Dreibladt in 1907, as a pseudoadenoameloblastoma.[3,4] Harbitz[5,6] in 1915 described it as a cystic adamantinoma. In 1948 Stafne[3,7] considered it a distinct entity. Unal et al[1,8]produced a list containing all nomenclatures for AOT reported in the literature. The tumour was named as adenoameloblastoma, adenoameloblastic odontoma, ameloblastic adenomatoid tumor, epithelial tumour associated with developmental cysts, adamantinoma, pseudoadenomatous ameloblastoma, epithelioma adamantinum or teratomatous odontoma before being currently defined as AOT. Philipsen and Birn[5,9]proposed the widely accepted and currently used name Adenomatoid Odontogenic Tumour in 1969, a term that was adopted by the first edition of WHO classification of odontogenic tumours in 1971.

The tumour appears as an intra- extra oral swelling in the maxilla and is sometimes referred to as " Two-thirds tumour" because it occurs in the maxilla in about 2/3 cases, about 2/3 cases occurs in young females, 2/3 cases are associated with an unerupted tooth and 2/3 of the affected teeth are canines[5,10]. According to Philipsen and Reichart[1,3,11,12], AOT appears in three clinico-topographic variants: the follicular type (accounting for 73% of cases), which has a central lesion associated with an embedded tooth; the extrafollicular type (24% of cases), which has a central lesion and no connection with the tooth; and the peripheral variety (3% of cases). As histogenesis of AOT is still uncertain, there has been a long debate as to whether it represents anomalous hamartomatous growth, or is a true benign neoplasm[5].

This paper presents an extrafollicular variant of adenomatoid odontogenic tumour, provides a refresher for general dental practitioner about the diagnostic aspects of this tumour and discusses the various presentations of this lesion.

CASE REPORT

An 18year old female reported with a painless swelling of anterior maxilla since approximately 18 months. Clinical examination revealed a well circumscribed firm growth in the buccal and palatal areas of gingiva extending from the distal surface of left maxillary lateral incisor to mesial aspect of canine displacing the teeth away from each other (Figure 1). Aspiration was negative and radiographic examination revealed a radiolucent lesion between lateral incisor and canine separating the two teeth without any root resorption (Figure 2). No evidence of any impacted tooth on the radiograph or any teeth missing on clinical examination was found. Based upon the clinical differential diagnosis of central giant cell granuloma or central ossifying fibroma, the lesion was enucleated under local anaesthesia, sent for histopathological examination and diagnosis. Surgical exposure of the lesion revealed cortical perforation and destruction of buccal cortical

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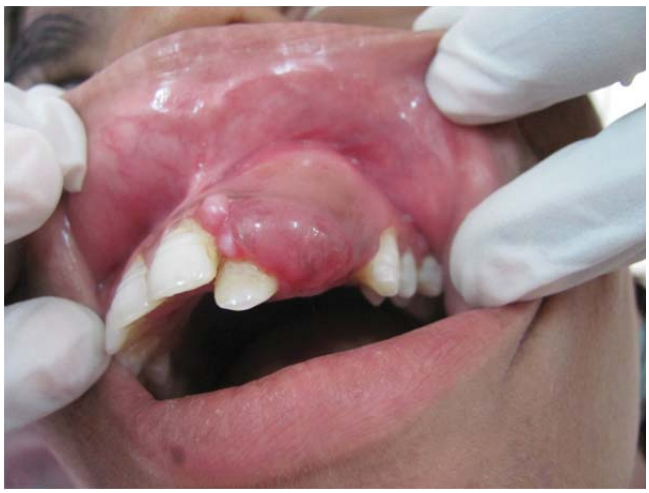

Fig 1 Preop Photograph showing the lesion

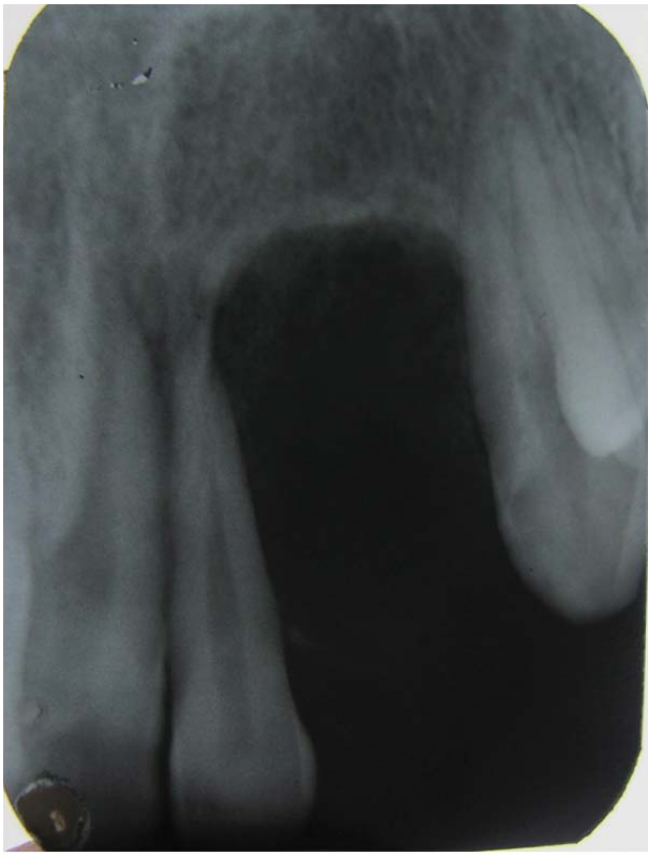

Fig 2. Preoperative Intraoral Periapical radiograph of the patient showing the extent of the lesion

plate. The teeth adjacent to the lesion were extracted owing to the bone loss distal and mesial to lateral incisor and canine respectively. The patient was followed for 25 months without any recurrence.

Grossly, the lesion was found to be a well-defined mass 25 X 15 mm in size surrounded by a well formed fibrous capsule (Figure 3). Histological examination showed numerous proliferation of cuboidal to columnar cells arranged in sheets and in some areas whorls, strands or cords of 1-2 cell thickness scattered

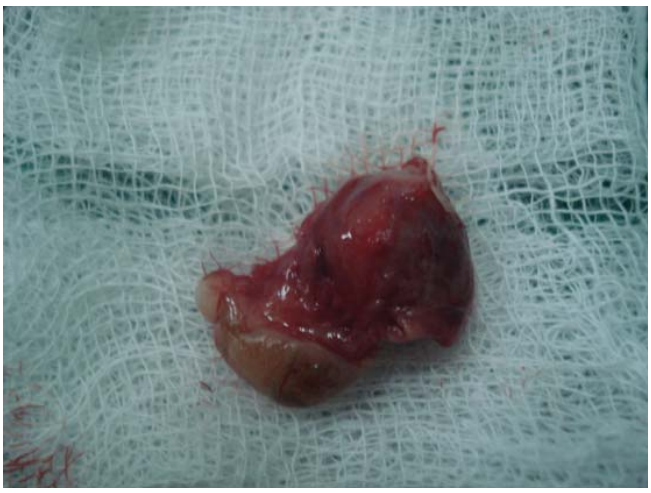

Fig 3. Excised tumour mass

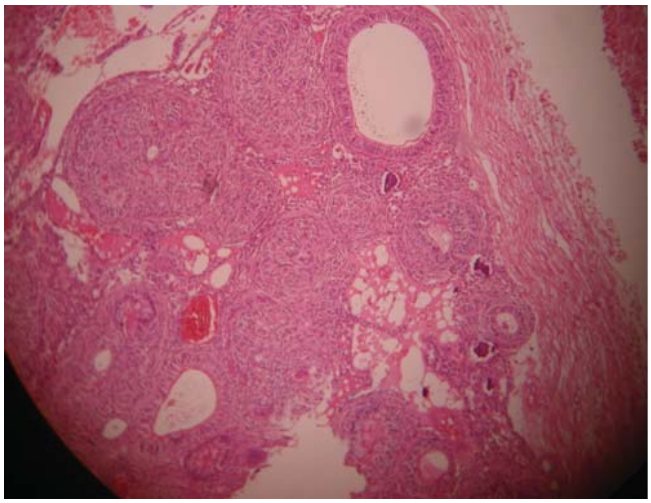

Figure 4. Histopathology of the lesion showing proliferation of cuboidal to columnar cells scattered in scanty connective tissue stroma

in scanty connective tissue stroma. Varying amounts of duct like structures were also seen with lumen of varying sizes lined by cuboidal to columnar cells with polarized nuclei located away from the lumen. Most of the lumen appeared to be empty with no eosinophilic material. Small amounts of dystrophic calcifications were seen between the ductal structures especially in the periphery of the lesion. Rest of the connective tissue stroma was loose and comprised of blood vessels and extravasated blood in some areas (Figure 4).

Discussion

AOT is a rare, slow growing, benign, odontogenic, epithelial tumours which usually arise in the second or third decade, the mean age being 13.2 years (range 3 until 28 years) and the female : male ratio being 2.3 : 1. AOT is usually located in the anterior maxilla (maxilla : mandible = 2.6 : 1); it produces a slow growing swelling without pain. The tumour growth may cause displacement of teeth

rather than root resorption. AOTs comprise only 0.1% of tumors and cysts of the jaws. Philipsen et al[1] reported that theyaccount for 3 to 7 % of all odontogenic tumors, 1.2% in Caucasian and 9% in black African patients. Information regarding incidence and prevalence of individual odontogenic tumors is still not available[13].

The origin of AOT is controversial[5,11,12,16]. Some support the idea that the lesion is a developmental outgrowth or hamartoma while others consider it to be neoplastic growth of odontogenic epithelium. The 1971 WHO classification stated: "It is generally believed that the lesion is not a neoplasm"[5]. However, Glickman et al,[5] concluded that "such a controversy is irresolvable because sound arguments can be advanced in favour of and against both hypotheses. The arguments are based on personal bias rather than on scientific evidence"

Due to its varied clinical and radiographic presentations, preoperatively, AOTs have been diagnosed as various types of disease[13]. Radiographically they frequently resemble dentigerous or follicular cysts. The radiolucency associated with AOT may extend more apically than that of a dentigerous cyst as seen in the present case. Irregular root resorption is seldom reported[5]. The varied picture of this tumour often confuses the practitioner thus misguiding them.

Clinical features generally focus on complaints regarding a missing tooth. The tumours are upto 1.5 to 3 cm, but larger lesions have been reported in the literature[13]. However, the rare peripheral variant occurs primarily in the gingival tissue of tooth-bearing areas[1]. They usually appear unilocular radiographically, but a few cases of multilocular radiolucency have been reported. Multiple AOT-like jawbone lesions were reported in a case by Larsson[13, 14]. AOT lesions may often appear completely radiolucent; however, they contain fine specks of dystrophic calcifications or tooth material like enamel, dentin, enamel and dentin, cementum, dentin and cementum, a feature differentiating AOT from dentigerous cysts[13]. In the present case, only dystrophic calcification was seen.

Both types of central intraosseous tumours produce a corticated radiolucency, sometimes with radiopaque specks. The follicular type is usually initially diagnosed as a dentigerous or a follicular cyst. According to Philipsen et al.[3,11], the extrafollicular type usually presents as a unilocular, well-defined radiolucency found between, above or superimposed on the roots of erupted teeth and often resembling a residual, radicular, globulomaxillary or lateral periodontal cyst. The peripheral type usually presents as a gingival swelling, located palatally or lingually relative to the involved tooth. Dare et al1,[15] found that intraoral periapical radiographs allow perception of the radiopacities in AOT as discrete focii having a flocculent pattern within radiolucency even with minimal calcified deposits while panoramic often do not. Those calcified deposits are seen in approximately 78% of AOT. Jham et al[13,17] reported a case of AOT arising from the periodontal ligament.

Table 1. Unusual findings seen in our case related to typical features of an adenomatoid odontogenic tumour (AOT)

Typical features of most AOT	Unusual findings in our case
Cortical penetration rare[18]	Cortical perforation seen
Unilocular radiolucency associated with an impacted unerupted tooth[18]	Not associated with impacted or or unerupted teeth

Since AOT is a benign tumor that presents with a non-aggressive biologic behavior, progressive growth, small frequency of recurrence, absence of invasion, and the frequent presence of a connective tissue capsule, the treatment should consist of enucleation and curettage[19]. If the follicle of the tooth is found intact, it can be easily separated from the tumour; it may be possible to remove the lesion while leaving the teeth in place, as described by Toida and others[3]. For periodontal intrabony defects caused by AOT, guided tissue regeneration with membrane technique is suggested after complete removal of the tumor1. Enucleation and curettage is the most common treatment modality for this tumour and recurrence is extremely rare. The prognosis is excellent.

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