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An unusual site of adenomatoidodontogenic tumor: a rare case report

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Abstract: Adenomatoid odontogenic tumour (AOT) is an uncommon tumour of odontogenic origin with relative frequency of 2.2-7.1%.AOT is a relatively rare and is exclusively of odontogenic epithelium origin. It is a benign, painless, noninvasive, and slow-growing lesion, often misdiagnosed as an odontogenic cyst on clinical examination. It is predominantly seen in young females in anterior maxilla, associated with unerupted canine. Here we are reporting a rare case of AOT in a 15-year-old female patient in the in mandible parasymphysis region. This paper highlights clinical, radiographic, histopathologic features andmanagement of the adenomatoid odontogenic tumor.
Keywords: adenoameloblastoma, ameloblastic adenomatoid tumor, Mandible; Odontogenic Tumors.

INTRODUTION

Adenomatoid odontogenic tumor (AOT) is a unique hamartomatous benign distinctively recognized odontogenic tumor .it may be located centrally or peripherally in the jaws. A large number of literatures is available which focus this lesion based on its clinical, radiological, histological and epidemiological characteristics[1]. Usualluy a variety of terms have been coined to describe this lesion of which the adenoameloblastoma was in common use for many years because the tumor is considered a one type of histological variant of ameloblastoma. It was first described by Dreybladt in 1907. The new generally accepted and very popularly used nomenclature of AOT is introduced by Philipsen and Birn in 1969. Some authors review AOTs as being truly benign, non-aggressive, non-invasive neoplasms in nature, whereas the secondary school of thought sees as developmental hamartomatous odontogenic growths. AOT occurs in intraosseous and peripheral forms [2]. the intrabony variants comprise the follicular and extrafollicular types. Radiographically the central lesions shows well demarcated perifollicular radiolucency with radiopaque focci often associated with an impacted tooth [3].

This report describes an AOT in the mandible; illustrating the clinical, radiological and histopathological feature.

CASE REPORT

A 15yr old female patient reported to the Department of Oral Medicine, Dr. Ziauddin Ahmad Dental College complaining of a swelling in the lower right jaw region. The patient reported that she noticed the swelling 2 months back, which was painless and gradually increased and attained the present size.

There was visible swelling on the right side of the mandible with mild facial asymmetry (Figure 1). The skin over the swelling and the surrounding area appeared normal and the margins were diffuse. On palpation, there was mild local rise in temperature, but the swelling was tender, especially in the anterior region

Intraoral Examination revealed a swelling which was reddish to pink in color and was approximately 2cm × 1cm in size. The swelling was obliterating the buccal vestibulein region of the first permanent right mandible molar (Fig-2). The swelling was bony hard in consistency and mildly tender on palpation. Hard tissue status revealed missing first premolar in concerned region.

Panoramicradiograph revealed a well defined radiolucent lesion with corticated border present in the body of the mandible of the right side extendingto adjacent canine and second premolar region .The lesion also extended inferiorlytill the lower border of mandible and seemed to be involving the inferior alveolar canal. The impacted first premolar adjacent to the lower border of mandible was also noted. A few radiopaque flecks of about 1-2 mm in the lesion were also noted. The lesion also resulted in displacement of roots of the canine and second premolar mesially and distally respectively (Fig-3).

On the basis of the clinical and radiographic findings, the differential diagnosis of adenomatoid

odontogenic tumour, ameloblastic fibrous odontoma, calcifying odontogenic cyst, calcifying epithelial odontogenic tumour and infected dentigerous cyst were considered.

Histopathological examination revealed cuboidal to columnar cells arranged in the form of nests and rosettes with minimal stromal connective tissue. Solid areas duct-like pattern, whorled arrangement of cells, and tubular appearance was evident. Convoluted structures were noted and at the periphery of the lesion tumor cells were arranged in a strand-like configuration. Few cells were also arranged in a plexiform pattern and cribriform areas were also seen. Some amount of calcification, eosinophilic material, and leisgang ring formation was also observed (Figure 4).

The histopathological report confirmed the diagnosis of adenomatoid odontogenic tumour.

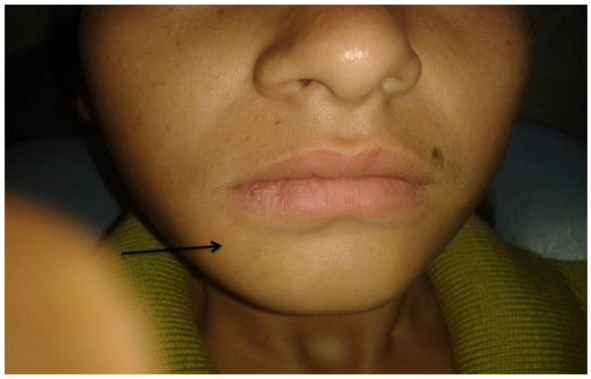

Fig-1: extraoral photograph showing a swelling in the right side of chin

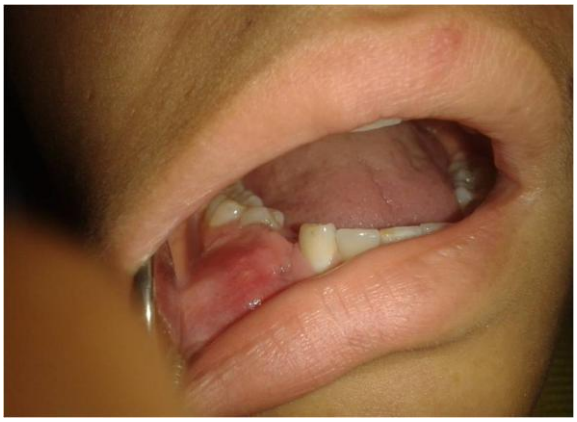

Fig-2: Intraoral photograph shows swelling in the anterior mandible

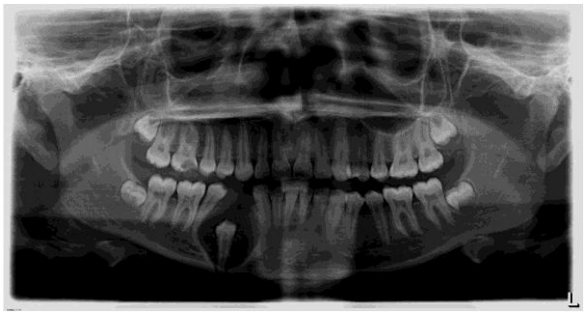

Fig-3: orthopantogram revealed well defined radiolucency radiolucency with radiopaque flecks associated with an impacted tooth in anterior mandible

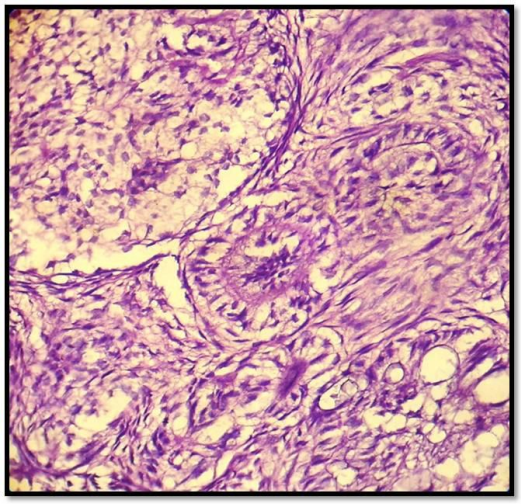

Fig-4: H/E section shows areas of odontogenic epithelium arranged in duct like and rosette pattern, fibrous stroma of spindle cells and 3-4 layer thick lining of cuboidal cells present

DISCUSSION

Adenomatoid odontogenic tumour is a relatively a slowly growing lesion, mostly seen in the anterior maxilla (ratio of cases 2:1 relative to mandible) of young females. The female to male ratio for all age groups and all variants is close to 2:1.10. the size of tumor usually lies in 1–3 cm in minimum to greatest diameter[4].

Usually the lesions are asymptomatic, but central type lesion may results in cortical expansion of adjacent bone. The teeth in question are commonly impacted resulting in displacement of adjacent teeth [5]. Our reported case also shows all the typical presentations of AOT in a 15-year-old female patient but with a rare location in the anterior mandible.

The origin of adenomatoid odontogenic tumours is still controversial. Some thought that its originate from the lining of odontogenic epithelium of a dentigerous cyst [6]. In addition to the anterior maxilla, the tumour has been rarely reported in other areas of the jaw, like the angle of the mandible. Therefore, dental

laminar remnants may likely to represent the progenitor cells for this unique benign odontogenic tumour. According to this hypothesis, the lesion grows (sometimes while forming a cystic pace) next to or involving a nearby dental follicle, leading to a theory called the "envelopmental theory" [7]. In the case reported here, the lesion surrounded a fully formed premolar tooth, which suggests "envelopmental" pathogenesis. Remarkably, all variants of AOT show identical histology. The histological typing of the WHO defined the AOT as a tumor of odontogenic epithelium with duct-like structures and with varying degrees of inductive change in the connective tissue. The tumor may be seen as partly cystic type, and in some cases the solid lesion may be present as well defined masses in the wall of a large cyst [8]. Moreover, eosinophilic, uncalcified, amorphous material can be found and are called "tumor droplets". Most tumor droplets show the electron-dense plaques, while some tumor droplets may show a homogenous matrix whereas [9].

Radiographically they usually appear as unilocular lesion, which usually contain fine calcifications with or without root resorption [10]. This appearance must be differentiated from various types of disease, such as calcifying odontogenic tumor or cysts. The differential diagnosis can also be made with ameloblastoma, ameloblastic fibroma and ameloblastic fibro-odontoma.

The tumor is well-encapsulated and shows an identical benign behavior. Therefore, conservative surgical enucleation produces an excellent outcome with good prognosis, with less chances of recurrence [11].

CONCLUSION

Odontogenic tumors including the so-called AOT comprise a unique heterogeneous group of lesions that ranges from hamartomas nature to benign and malignant neoplasms of variable aggressiveness behaviour. Our case report supports the general description of adenomatoid odontogenic tumor in the previousstudies. The AOT in the mandible is very rare but characteristic feature of this help in diagnosing the lesion. Careful diagnosis and adequate interpretation of clinical, radiological finding and gold standard histopathological evaluation help in arriving at correct diagnosis.

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