

Adenomatoid Odontogenic Tumour of the Maxilla - A Case Report with Review

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ABSTRACT

Adenomatoid odontogenic tumour is a relatively uncommon odontogenic tumour first described by Steensland in 1905. Adenomatoid odontogenic tumour accounts for about 3-7% of all odontogenic tumours, predominantly found in young female patients, located more often in maxilla associated with an unerupted permanent tooth. The tumour is often misdiagnosed as an odontogenic cyst. AOT frequently resembles other odontogenic lesions such as dentigerous cysts or ameloblastomas. Therefore, it should be distinguished from the more common lesions of odontogenic origin in routine dental examinations. We present a rare case report of 59-year-old female patient of AOT occurring in the maxilla.

Keywords: Adenomatoid odontogenic tumour, Neoplasm, Duct like structures, Extra follicular

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INTRODUCTION

Adenomatoid Odontogenic Tumour (AOT) is a relatively uncommon, benign and slow growing tumour which is often misdiagnosed as an odontogenic cyst (1,2). AOT accounts for 3-7% of all the odontogenic tumours (3). It is predominantly found in young and female patients, is located more often in the anterior maxilla in most of the cases and is associated with an unerupted permanent tooth, usually the lateral and canines in the 2nd decade (4). It often causes the expansion of the surrounding bone and the displacement of the adjacent teeth (5). However, the slow growing and painless nature of the lesion may cause the patients to tolerate the swelling for years until it produces an obvious deformity (6). A variety of terms have been used to describe this tumour, like adenoameloblastoma, ameloblastic adenomatoid tumour, adamantinoma, epithelioma adamantinum or teratomatous odontoma, which is currently called as AOT (7,8). In 1999, Philipsen and Riechart presented a review based on reports which were published until

1997, which showed some interesting aspects regarding the epidemiological figures of this tumour (9). Since then, numerous case reports of AOT have been published. In this case report, we present a 59-year-old female patient of AOT occurring in the maxilla.

CASE REPORT

A 59-year-old female patient reported to department of oral medicine and radiology with a chief complaint of pain and swelling in the left side of the upper front region for past 6 months. (Figure 1) She gives history of trauma



Figure 1. Profile picture

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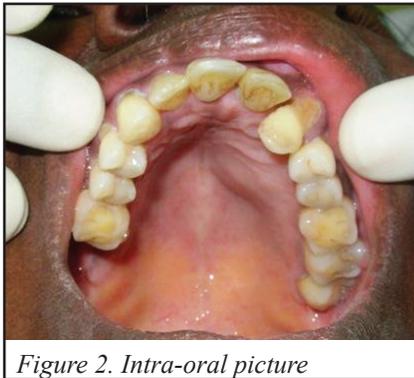


Figure 2. Intra-oral picture

2 years back, for which she visited a local doctor and taken medications. Now for last 10 days she noticed the swelling, which is slowly increasing in size. On examination, extraorally facial asymmetry seen with a single diffused swelling evident on the left side of the face, roughly measuring about 6x5 cm in size extending superiorly 2 cm below the lower eye lid and extends up to the angle of the mouth inferiorly. Medially it crosses the midline and obliterating the nasolabial fold and philtrum. (Figure 2) The swelling is mildly tender, firm in consistency. Intraorally, on examination 22 is missing, 23, 24, and 25 were grade III mobile and cervical abaration were present and 23 is palatally placed. A single diffused swelling evident on the labial aspect in relation to 21 to 26 regions, roughly measuring about 3x2 cm in size. The swelling extends anteriorly from the midline to the distal aspect of 26 region posteriorly. Superiorly, obliterating the labial vestibule, inferiorly, extending up to the gingival margin of 21 to 26 with no secondary changes. On palpation the swelling is mildly tender, Soft in consistency in relation to 21, 25, 26 region

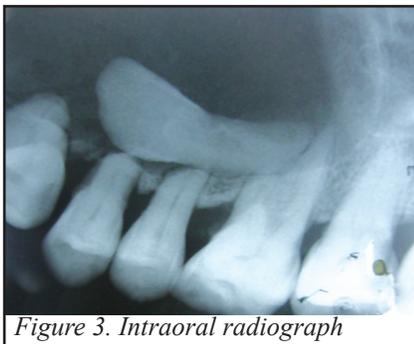


Figure 3. Intraoral radiograph



Figure 4. Surgical specimen

and firm in consistency in relation to 23, 24 region. Eggshell crackling felt in relation to 23 regions. Vitality test were performed with gutta-percha point shows 11- Delayed response, 12, 13, 24, 25 - Response, 21, 23-No response.

The periapical radiograph showed a horizontally placed impacted 22, with diffuse Radiolucency evident in relation to the periapical aspect of 23,24,25,26 with root resorption. (Figure 3) Maxillary occlusal view shows diffuse Radiolucency involving the left side maxillary region roughly measuring about 4x2 cm in size, extending from 21 to 26 regions. Based on history and clinical presentation, a provisional diagnosis of AOT was made. The complete hemogram of the patient was within normal limits. Fine needle Aspiration cytology done 4ml of straw-coloured cystic fluid obtained and sent for biochemical analysis, the protein analysis report was 6.3 gm%. The lesion was totally enucleated, and the tissue was sent for histopathological evaluation. (Figure 4) Histopathological examination shows a thick fibrous capsule enclosing spindle shaped cells arranged in whorl-like pattern and

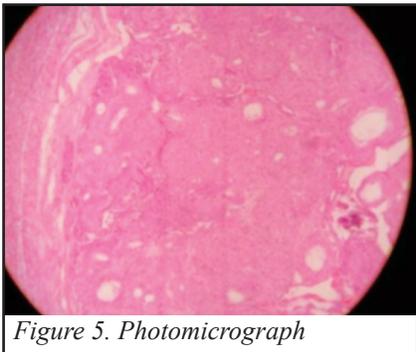


Figure 5. Photomicrograph

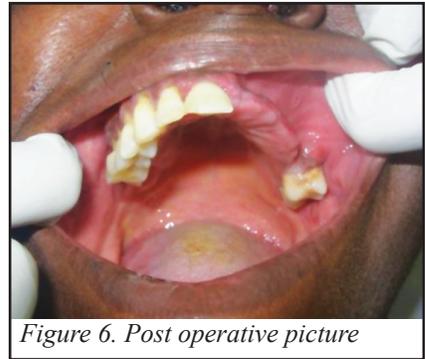


Figure 6. Post operative picture

numerous duct like structures lined by cuboidal cells enclosing eosinophilic amyloid like material. Basophilic calcifications are also seen. Suggestive of Adenomatoid odontogenic tumour (Figure 5) Patient was reviewed once in 3 months for 1 year. (Figure 6)

DISCUSSION

The AOT is a benign non-invasive rare odontogenic tumour, showing slow growth which causes jaw swelling (10). There is a slight female over male predilection which is almost 2:1 and it appears most often in the second decade of life (11). In this case, the lesions were typically asymptomatic, but they could cause the cortical expansion and the displacement of the adjacent teeth. The origin of the AOT is controversial, but as it arises in the tooth bearing area, it is thought to arise from the odontogenic epithelium i.e. the cell rests of malessez (12). The tumour has three clinicopathological variants, namely intraosseous follicular, intraosseous extra follicular, and peripheral. The follicular type is a central intrabony lesion associated with an unerupted tooth, which accounts for about 73% of all the AOT cases. The extra follicular type is also an intra-osseous lesion but not related with an impacted tooth, which accounts for about (24%) of all the AOT cases and the peripheral variant is a rare form and is attached to the gingival structures and 18 well documented cases reported, which accounts for about (3%) (13).

Most of the intra-osseous variants are discovered on routine radiographic

examination. However delayed eruption of the tooth, slow growing, bone expansion, with or without development of the teeth, asymmetrical facial swelling can be the presenting signs (14). The tumours are usually in the dimensions of 1.5 to 3 cms radiographically, they appear as unilocular and may contain fine calcifications and an impacted tooth with the displacement of the adjacent roots (15).

WHO (16, 17) has described the histologic features of the tumour as follows, "A tumor of odontogenic epithelium with duct like structures and with varying degree of inductive changes in the connective tissue. The tumor may be partly cystic and in some cases the solid lesion may be present only as masses in the wall of a large cyst. It is generally believed that the lesion is not a neoplasm." The histologic appearance of all variants is identical and exhibits remarkable consistency. (9,17,18) At low magnification 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. Between the epithelial cells of the nodules and in the centre of the rosette-like configuration is found eosinophilic amorphous material, often described as tumour deposits. Conspicuous within the cellular areas are structures of tubular or duct-like appearance. A third characteristic cellular pattern consists of 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 (thus the suggestion of a combination of calcifying epithelial odontogenic tumour and adenomatoid odontogenic tumour) (18). Immunohistochemical studies of the lesion suggest expression of keratin and vimentin in the tumour cells at the periphery of the ductal, tubular or whorled structures.

Since all variants show identical benign biological behaviour and almost all are encapsulated, conservative surgical enucleation or curettage is the treatment of choice. Recurrence has been reported in very few cases (9).

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