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Case Report

Case reports of a typical extrafollicular adenomatoid odontogenic tumor of maxilla

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ABSTRACT

Adenomatoid odontogenic tumor is a rare benign odontogenic tumor comprising of 3% to 7% of all Odontogenic tumors. It is usually asymptomatic, well circumscribed, slow growing tumor associated often with impacted canine and discovered on routine radiographs. Microscopically this lesion shows a variety of patterns forming sheets, strands, nest or rosette-like structures with ductal pattern with central space surrounded by single layer of cuboidal to columnar epithelial cells with a rim of eosinophilic coagulum which is the characteristic feature of these tumors. In this article we present two extrafollicular cases of this rare tumor occurring in a male and a female patient in the maxillary anterior region with both symptomatic and asymptomatic presentation. Both the tumors were not associated with any impacted tooth. Adenomatiod Odontogenic tumor was confirmed based on histopathology.

Keywords: Adenomatoid odontogenic tumor, Benign, Hamartoma, Histomorphological patterns

INTRODUCTION

Philipsen and Birn first introduced the term Adenomatoid Odontogenic tumor (AOT) in 1969, later on adopted by World Health Organization (WHO) in 1971.[1,2] However it was first described by Ghosh as an adamantinoma of the maxilla and Stafne recognized it as distinct entity.[3] WHO (2005) defined it as "AOT is composed of odontogenic epithelium in a variety of histoarchitectural patterns, embedded in a mature connective tissue stroma and characterized by slow but progressive growth".[4,5] Due to its non-invasive and non aggressive nature, many authors believe it to be a benign neoplasm.[6] While few authors consider AOT as a hamartoma due to its limited size and lack of recurrence after complete removal.[2,7] Radiographically intrabony variants comprise of follicular and extrafollicuar types. Follicular type is unilocular and associated with an unerupted tooth resembling a dentigerous cyst. Extrafollicular type is not associated with unerupted tooth, but most commonly seen around erupted permanent tooth.[8] Treatment of AOT involves conservative surgical enucleation. Recurrence is rare.[9]

CASE REPORT

Case 1

A 38 years old female patient was referred to the department of oral pathology and microbiology, GSL

Dental College and Hospital, Rajahmundry with a chief complaint of swelling on the right maxillary region. Patient noticed the swelling one month back and was asymptomatic. There was no history of dental caries or pus discharge and tooth mobility. No relevant medical history. On further examination mild extraoral bony swelling was noticed in the right maxillary canine region. Intraoral periapical radiograph (Figure 1) and Orthopantomogram (Figure 2) showed a radiolucent lesion between the roots of right maxillary lateral incisor and canine and the roots were displaced.

Figure 1: IOPA showing radiolucency between lateral incisor and canine.

Figure 2: OPG showing radiolucency between 12 and 13 with displacement of roots.

Excisional biopsy of the lesion revealed a well encapsulated cystic lesion. Macroscopic examination of the cut lesion showed white gritty material inside the cystic cavity, with a thick fibrous capsule (Figure 3).

Microscopically epithelial cells arranged in the form of whorls or rosettes and ducts lined by a single layer of cuboidal to columnar cells were seen. Few areas showed basaloid epithelial cells resembling cell rests of dental lamina (Figure 4). The fibrous capsule showed variable thickness (Figure 5).

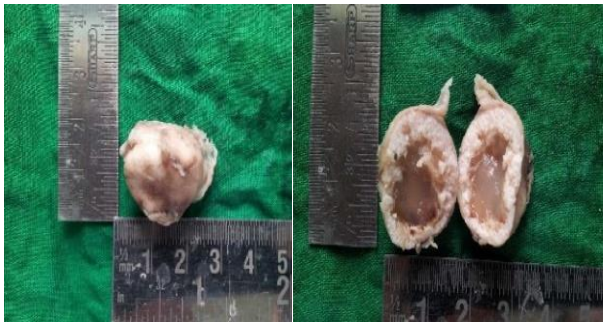

Figure 3: Well encapsulated lesion with cystic and thick fibrous capsule containing white gritty substance.

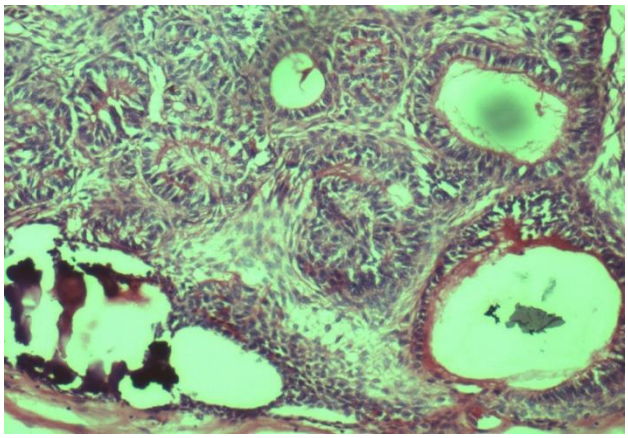

Figure 4: Microscopically, cuboidal to columnar cells arranged in ductal pattern with a rim of eosinophilic coagulum are seen under high power view.

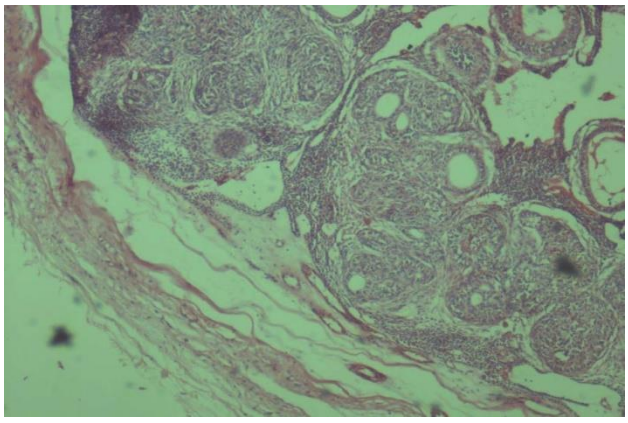

Figure 5: Low power view showing a thick fibrous capsule with ductal pattern.

Case 2

A 21 years old male patient reported to the GSL Dental College and Hospital, Rajahmundry with swelling and pain in the maxillary anterior region since one week and was gradually increasing in size. On extraoral examination a mild swelling was noticed below the ala of nose extending up to the corner of the mouth (Figure 6).

Figure 6: Mild swelling seen extraorally below the ala of nose up to the corner of the mouth.

Intraorally a 2 x 2 cm swelling was noticed between maxillary left lateral incisor and canine. It was soft, fluctuant and tender on palpation (Figure 7).

Figure 7: Obliteration of vestibule with swelling extending from central incisor to canine.

Radiographic examination revealed a well demarcated radiolucent area between 22 and 23. Roots of 22 and 23 were displaced (Figure 8).

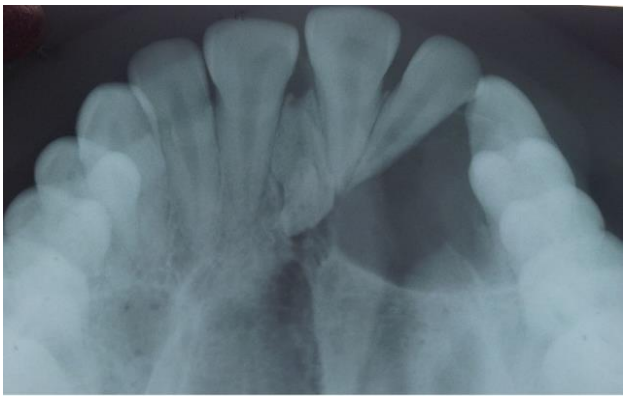

Figure 8: Radiographically a well demarcated radiolucent area between 22 and 23 is noticed.

Provisional diagnosis of periapical cyst or lateral periodontal cyst was considered. Excisional biopsy revealed a well encapsulated cystic lesion with thin watery fluid was found (Figure 9).

Figure 9: Biopsy revealed a well encapsulated cystic lesion with thin watery fluid.

Histopathological examination revealed ductal pattern of cuboidal or columnar cells (Figure 10).

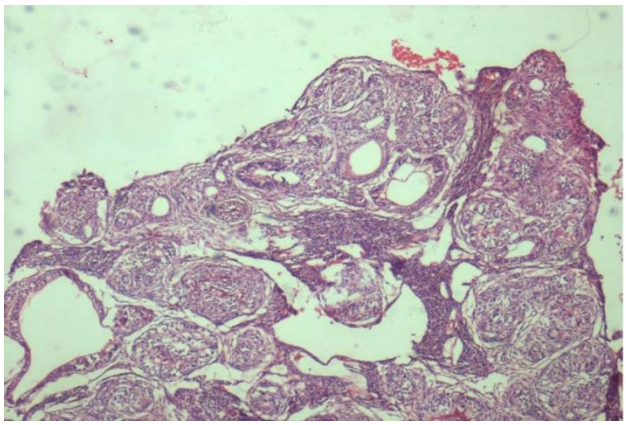

Figure 10: Microscopic examination revealed ductal pattern of cuboidal or columnar cells.

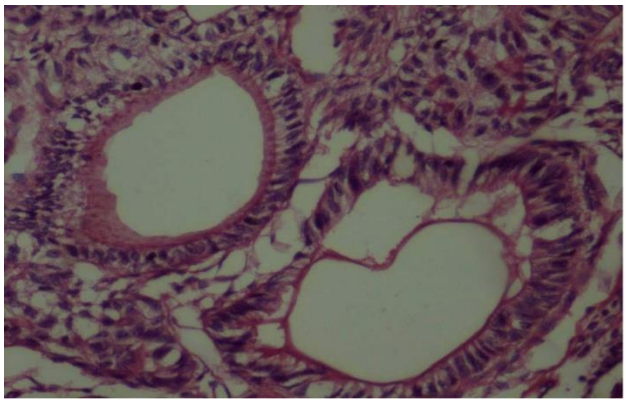

Figure 11: Under high power,spindle cells are seen arranged in the form of sheets and anastomizing cords in an eosinophilic matrix.

Spindle cells were seen arranged in the form of sheets and anastomizing cords in an eosinophilic matrix. Few

areas showed stellate reticulum like spindle cells present between the cell rich nodules (Figure 11). Based on histopathology AOT was confirmed.

DISCUSSION

AOT is an uncommon Odontogenic tumor commonly occurring in the maxillary anterior region. It was first reported by Steensland as 'epithelioma admantinoma'.[10] AOT is a slow growing, asymptomatic lesion, seen in young females however few cases have been reported males also and is common in the second decade of life. It is commonly known as a "two-thirds tumor" as 2/3rd of these tumors are associated with females, 2/3rds of the cases occur in Maxilla, and 2/3 of the cases are associated with an unerupted tooth.[11] Clinicopathologically three variants have been identified namely intraosseous follicular, intraosseous extrafollicular and peripheral. Follicular variant is more common type of AOT compared to other two variants. Follicular variant should be differentiated from dentigerous cyst. Dentigerous cyst encloses only the coronal portion of an impacted tooth whereas AOT shows radiolucency surrounding both the coronal and radicular aspects of the involved tooth. Displacement of the teeth associated with tumor is more common.[12] Radiographically AOT presents as a well demarcated, unilocular radiolucency that generally exhibits a smooth or corticated border. Most lesions are around 1 to 3 cm in greatest diameter and associated with impacted tooth.[13] Divergence of roots and displacement of teeth occurs more frequently than root resorption.[14]

AOT exhibits diverse features in histopathology. The tumor shows proliferation of spindle, cuboidal and columnar cells in variety of patterns. As many as 20 different histological patterns of this tumor have been described (Table 1). Usually duct like structures, with eosinophilic material or calcifications with a thick fibrous capsule are seen. Occasionally round or polygonal epithelial cells may dominate the tissue between the cell rich nodules.[15] Small amounts of eosinophilic material or calcifications also may be present between these cells.[16]

Siar et al has described a cystic variant of AOT with mural lining.[17] Similarly Philipsen et al has noted CEOT like areas, cribriform pattern type, interlacing strands of cells, nest, rosette, trabecular and tubular like patterns.[18] Fredrich et al., described a case of AOT with net like proliferations by peripheral smaller cells.[19] Ribbon, ring, whorled spheroidal pattern and sieve-like, solid nodules type pattern where discussed by Garg et al and Takahashi et al respectively.[20,21]

Although the location of AOT was typical, both cases described in the present article were of extrafollicular variant types as the lesion was seen between roots of Maxillary lateral incisor and canine, with displacement of the roots. The occurrence of this tumor in male patient in the third decade with symptoms of pain is quite uncommon.

AOT origin is considered controversial, while some believe it is from the odontogenic epithelium of the dentigerous cyst and others consider it to be derived from epithelial remnants of the dental lamina complex system.[22] Chen et al. has stated that AOT derived from dentigerous cyst and suggested the term "hybrid variant".[23]

Immunohistochemical analysis usually indicates that AOT is a noninvasive tumor that never infiltrates surrounding normal tissues. These tumors exhibit weaker expressions of p53 and MDM2 indicating their less aggressive behavior.[24,25] Enamel proteins including amelogenin, ameloblastin, and amelotin, as well as TGF-β/SMADs, are more intensely expressed in AOT.[26,27]

CONCLUSION

Therefore, AOT can present themselves with variations in size, gender and location, sometimes mimicking other odontogenic tumors. However, irrespective of the pattern the biological behavior of the tumor has never changed unlike that of the other tumors. Conservative surgical enucleation is the treatment of choice. Recurrence of AOT is exceptionally rare with only few cases reported indicating excellent prognosis.

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