Review Article

Adenomatoid Odontogenic Tumor (AOT): An Overview

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ABSTRACT

Adenomatoid Odontogenic Tumor (AOT) is benign epithelial lesion of odontogenic origin derived from complex system of dental lamina remnants. The lesion is well known for various clinical and histopathological appearances and so it is has been given the title 'master of disguise'. It is well-recognized as a leisurely growing benign tumor which is not rare now as it was previously reported. This lesion is grouped into three variants of which the most common variant is follicular type which often presents as dentigerous cyst clinically because it surrounds an impacted tooth. We report an AOT of mandible in a 14-year-old male who reported to our department and was provisionally diagnosed as unicystic ameloblastoma or dentigerous cyst, along with a review of literature on the tumor. Clinical, radiological, histopathological characteristics and treatment modality of the case have been stressed upon.

Keywords: Adenomatoid odontogenic tumor, benign tumor, follicular type

INTRODUCTION

The lesion now known as the adenomatoid odontogenic tumor, was documented as a separate entity by Stafne.[1] The earliest recorded case was in 1907 by Driebladt who described it as a pseudo-adenoma-adamantinoma. This tumor was assigned various names until Philipsen and Birn introduced the name 'Adenomatoid odontogenic tumor' (AOT). The histologic typing of WHO in 1971 introduced a definition for this lesion describing it as, "A tumour of odontogenic epithelium with duct-like structures and with varying degrees of inductive change in the connective tissue. The lesion 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".[2]

It is now a not so rare benign epithelial lesion derived from the remnants of dental lamina enveloping a developing dental follicle which is mostly solid and may be partly cystic.

AOT constitutes 2.7 to 7% of all odontogenic tumors.[3] It is primarily found in young females with a male:female ratio of 1:2, occuring largely in the maxilla in association with an impacted permanent tooth. For radiological diagnosis, the intraoral periapical radiograph holds more diagnostic value than panoramic. AOT exhibits resemblance to other odontogenic unilocular cystic lesions enclosing the unerupted teeth such as dentigerous cyst or ameloblastoma in its clinical and radiographic appearance. Histopathologically also, the lesion is sometimes found to contain a cystic component.

Based on the clinical and radiologic findings, AOT can be subdivided into different variants:[4] Central (or intraosseous) variants: 1) Follicular type which encompasses the crown

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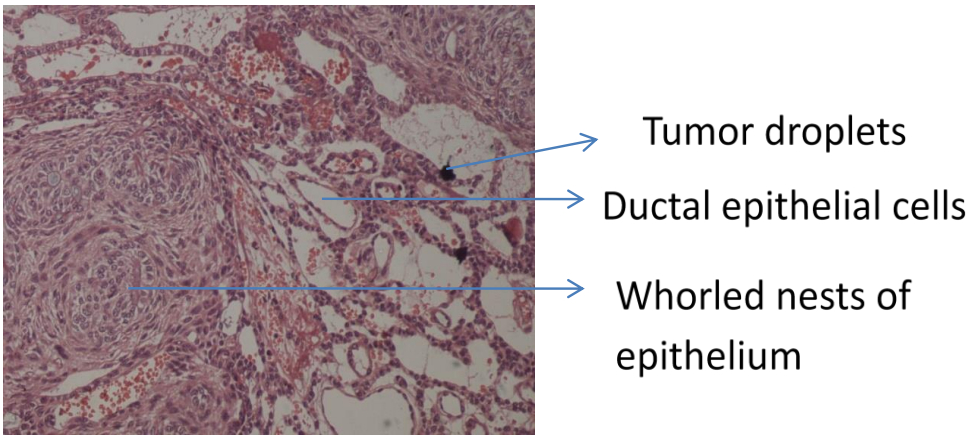

Figure 1: 200X microphotograph demonstrating columnar epithelial cells arranged in whorled nest pattern, ductal cells represented by clear cells lined by single layer of epithelia cells nuclei of which show changed polarity. Tumor droplets are seen.

of an unerupted tooth thereby, resembling a dentigerous cyst.

2) Extrafollicular type has no association with the crown of an unerupted tooth. The provisional diagnosis of this variant could be a 'residual', a "globulomaxillary" or a lateral periodontal cyst depending on the actual intraosseous localization of the lesion.

Peripheral (or extraosseous) variant with a resemblance to gingival fibroma or fibrous epulis.

MATERIAL AND METHODS

PubMed or Scopus indexed articles were included in this review. A total of 38 articles were taken after 1990 till 2019 and a review of the clinical, radiographic and microscopic features has been reviewed along with the treatment methods adopted by various authors and deviation from regular treatment methods if any has also been discussed.

Clinical features

The lesion usually presents as asymptomatic swelling which is slowly growing and often associated with an unerupted tooth. However, the rare peripheral variant occurs primarily in the gingival tissue of tooth-bearing areas.[5] Unerupted permanent canine are the teeth most often involved in AOTs.

Radiographic features

The radiographic findings of AOT frequently resemble other odontogenic lesions such as dentigerous cysts, calcifying odontogenic cysts, calcifying odontogenic tumors, globule-maxillary cysts, ameloblastomas, odontogenic keratocysts and periapical diseases.[6] Whereas the follicular variant shows a well-circumscribed unilocular radiolucency associated with the crown and often part of the root of an unerupted tooth, the radiolucency of the extrafollicular type is located between, above or superimposed upon the roots of erupted permanent teeth.[7] Displacement of teeth in the vicinity of tumor is a common occurrence. Resorption of roots may or may not be present. The peripheral lesions may show some erosion of the adjacent cortical bone.[4] Dare et al.[8]] found that intraoral periapical radiographs allow perception of the radiopacities in AOT as discrete foci having a flocculent pattern within radiolucency even with minimal calcified deposits which panoramic often do not. Those calcified deposits are seen in approximately 78% of AOT.[9] In addition, in one recently reported case MRI was useful to distinguish AOT from other lesions, even if it is difficult on periapical ordinary radiographies.[6]

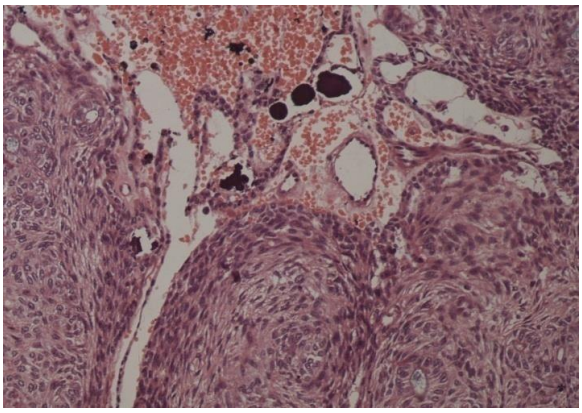

Figure 2: 200X Microphotograph demonstrating calcified deposits within the lesion.

Microscopic features

AOT is usually surrounded by a well-developed connective tissue capsule. It may present as a solid mass, a single large cystic space, or as numerous small cystic spaces. The tumor is composed of spindle-shaped or polygonal cells forming sheets and whorled masses in a scant connective tissue stroma [Fig. 1 & 2]. Between the epithelial cells as well as in the center of rosette-like structures is amorphous eosinophilia material. The characteristic duct-like structures is lined by a single row of columnar epithelial cells, the nuclei of which are polarized away from the central lumen. The lumen may be empty or contain amorphous eosinophilic

material. Dystrophic calcifications in varying amounts and in different forms are usually encountered in AOTs, within the lumen of duct-like structures, scattered among epithelial masses, or in the stroma.[4]

Treatment and prognosis

Conservative surgical enucleation is the treatment modality of choice. For large intrabony defects caused by AOT, bone grafts can be used or guided tissue regeneration with membrane technique can also be done or the small defects can be left to heal.

DISCUSSION

In 2005 WHO defined it as 'A tumor composed of odontogenic epithelium, presenting a variety of histoarchitectural patterns, embedded in mature connective tissue stroma and characterized by slow and progressive growth.'

Radiographically, it is mostly seen as a well-defined unilocular radiolucency sclerotic borders. On the basis of clinical and radiologic features, the follicular type comprising a central lesion encompassing an unerupted tooth accounts for 70.80% of the cases; the extrafollicular type 26.9% of the cases whereas the peripheral variety forms 2.3% of the cases.[10] Both variants of central tumors present a corticated radiolucency, sometimes interspersed with radiopaque specks. It is the follicular type which usually is diagnosed as a dentigerous cyst or unicystic ameloblastoma initially. The extrafollicular type often resembles a residual, radicular, globulomaxillary or lateral periodontal cyst. The peripheral type presents as a gingival swelling, located palatally or lingually in relation with the involved tooth.

Epithelial tumor cell components in AOT have been described into three cell types.[11]

Cell type I: small compact cells in a solid nodule and pseudoglandular cells in a duct-like structure; Cell type II: peripheral elongated cells and spindle shaped cells in a cribriform pattern; and Cell type III: metaplastic squamous cells.

Calcifying epithelial odontogenic tumour (CEOT)-like areas are also found in some cases of AOT. AOT cases show positive results for AE1/AE3, 34bE12, CK5, CK14 and CK1. Vimentin is also expressed in some cases and this may suggest the pattern of varied phenotypical characteristics in certain areas of the tumour. The low recurrence after surgical treatment can be attributed to the low proliferative activity observed in cases with Ki-67 marker.[3]

The origin of this tumor is controversial. The dental lamina remnants could probably be the parent cells for this benign odontogenic tumor. According to this theory, the lesion grows adjacent to or into a nearby dental follicle

leading to the "envelopmental theory".[12] Santos et al.[13] reported a case of AOT developing in the fibrous capsule of the dentigerous cyst. Garcia Pola et al.[14,15] described the proliferation of an AOT in the epithelial borders of a dentigerous cyst. Cassiano Francisco Weege et al.[14] also reported a case of AOT associated with dentigerous cyst. The tumor associated with a dentigerous cyst is reported to involve not only the anterior maxilla but other areas of the jaw such as the angle of the mandible. The reason for this correlation between an odontogenic cyst and tumor can be attributed to the fact that neoplastic and hamartomatous changes can occur at any stage of odontogenesis. Iron-binding proteins and proteinase inhibitor may be involved in the pathogenesis of AOT.[16]

Various studies and literature support the fact that the maxillary arch is the predominant site of occurrence, being almost twice as frequent as that of the mandible. Giansanti et al. reported that 65% AOTs were seen in the maxilla and 35% in the mandible. In a recent retrospective study of 61 cases in Nigerian population, 55.8% cases were seen in maxilla whereas 32.8% cases were found in the anterior mandible.[17] Of the mandibular lesions, 69% were found in the anterior region, 27% in the premolar region, and a few in the molar region.[18] Mandibular AOTs are similar to maxilla in their clinical as well as radiographic pattern. Though, mandibular AOTs are a rare finding one should never ignore the possibility of finding it in the mandibular anterior region when there is an impacted canine associated with it.

Clinically and radiographically the lesion mimicks a unicystic ameloblastoma. These lesions can grow to considerable sizes also involving the lower border of the mandible. Owing to its benign behavior, slow growth and clear margins, as well as its low tendency to recur, the treatment of choice is enucleation and simple currettage, although in exceptional cases of large tumors, there is risk of bone fracture. Partial resection, en bloc of the mandible or maxilla has been indicated for such cases.[19] Additionally, the use of lyophilized bone and guided tissue regeneration is recommended in cases where surgical enucleation leaves a large exposed osseous cavity.[19] The resection of mandible in growing young patients is associated with plethora of complications such as loss of jaw bone support, deformity, dysfunction and psychological distress even after good reconstruction. Therefore, an alternative conservative surgical procedure "Dredging Method" which has been reported with good success rate in ameloblastoma[20] can also be tried for large, aggressive AOTs. This calls for more research in this area. Conservative surgical enucleation is the most suitable treatment option available for AOT as it has almost nil recurrence rate as evident in various studies demonstrating no recurrence over a period of time. Only three cases in Japanese patients are reported in which the recurrence of this tumor occurred.[21]

The histopathology report revealed typical features of adenomatoid odontogenic tumor, follicular type which usually establishes itself around the crown of unerupted, anterior teeth in young patients, being constituted of whorled nests of duct like epithelial cells together with areas of glandular or ductal patterns intermixed with occasional spherical calcifications. It is interesting to note that irrespective of the pattern, the biological behavior of the tumor never changed unlike that of the other tumors as confirmed by various studies on the biologic behavior of this tumor. Although it presents with varied histopathological patterns, its histology also has always remained distinct making it easy to diagnose this tumor. This calls attention for stressing more on the histomorpholgy of AOT and in knowing why none of these patterns have any effect on the biological behavior of these tumors. Aggressive variant of this tumor has also been reported in literature with 8 cases of this variant being reported till date.[22] The treatment in these cases remains more or less the same and depends on many factors like age of the patient and proximity to important anatomical structures.

CONCLUSION

Though AOT is rare in mandible, we can conclude that it is now slowly becoming less and less rare. They are easily diagnosed owing to their striking histopathology. Enucleation followed by surgical curettage remains the gold standard of treatment for these lesions.

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