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A Rare Case Report of Cystic Adenomatoid Odontogenic Tumour in the Mandibular Anterior Region

[1]Dr. Swati Choudhary, Reader, MDS, Department of Oral Pathology, ITS Dental College and Hospital, Greater Noida.

[2]Dr. Kehkashan Ehsan Azmi, Senior Lecturer, MDS, Department of Oral Pathology, ITS Dental College and Hospital, Greater Noida.

[3]Dr. Aditya Saxena, Professor, MDS, Department of Pedodontics, ITS Dental College and Hospital, Greater Noida.

[4]Dr. Deepesh Choudhary, Senior Lecturer, MDS, Department of Prosthodontics, ITS Dental College and Hospital, Greater Noida.

[5]Dr. Karan, Reader, MDS, Department of Oral Surgery, SBB Dental College.

Corresponding Author: Dr. Swati Choudhary, Reader, MDS, Department of Oral Pathology, ITS Dental College and Hospital, Greater Noida.

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Abstract

Adenomatoid Odontogenic Tumor (AOT) is renowned for its deceptive nature, often masquerading as other lesions due to its diverse clinical and histological manifestations. Typically found in the anterior maxilla, it commonly presents as a painless swelling associated with an impacted tooth. Radiographically, it appears as a well-defined, unilocular radiolucency, mimicking features seen in dentigerous cysts. However, AOT can also display cystic components histologically, further complicating its diagnosis. The present case involves a 20-year-old male patient having a clinical diagnosis of eitherameloblastoma or dentigerous cyst but on excisional histopathological analysis cystic AOT was confirmed.

Keywords: Adenomatoid Odontogenic Tumour; Impacted Tooth, Unilocular, Anterior Mandible, AOT.

Introduction

Adenomatoid Odontogenic Tumor (AOT) is a rare epithelial tumor, constituting around 3% of all odontogenic tumors. It was first reported by Steensl and in 1905 and initially termed as pseudo-adenoameloblastoma by Dreibladt in 1907.[i] There was debate regarding its classification, with some considering it a variant of ameloblastoma until Stafne in 1948 proposed it as a distinct entity. [iii, iv] However, it wasn't until 1969, when Philipsen and Birn introduced

the name 'adenomatoid odontogenic tumor', which was later adopted by the World Health Organization (WHO) in 1971.[iii] Prior to this, various terms such as adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantinum, and teratomatous odontoma were used to describe this lesion. Additionally, in 2003, Max and Stern coined the term 'adenomatoid odontogenic cyst'. [ii, iii]

The Adenomatoid Odontogenic Tumor (AOT) is characterized as a benign, painless, noninvasive, and slow-growing lesion that typically does not penetrate bone tissue. Clinically, it frequently masquerades as an odontogenic cyst. It typically presents as a swelling either within the oral cavity or externally in the maxilla. Interestingly, it's sometimes dubbed the 'two-third tumor' due to its prevalence: approximately two-thirds of cases occur in the maxilla, two-thirds affect young females, two-thirds are associated with an unerupted tooth, and two-thirds involve canine teeth. [v],[vi] The precise origin of AOT is still debated. However, it's believed to stem from odontogenic epithelium given its occurrence in tooth-bearing areas of the jaws, association with impacted teeth, and presence of components resembling those of the enamel organ, dental lamina, reduced enamel epithelium, or their remnants. [iv] The tumour 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. Though the definition states the lesion may have a cystic nature very few case reports have described the cystic lining. The objective of this article is to present and analyze a unique case of Cystic Adenomatoid Odontogenic Tumor (AOT) found in the mandible. It primarily focuses on scrutinizing the radiographic characteristics while correlating them with pathological findings.

Presentation of case

A 20-year-old male patient presented with a complaint of a pain and swelling on his right anterior region of the jaw since 15 days. Clinical examination revealed a well-defined hard unilateral swelling extending antero-posteriorly involving the gingiva from 83 to 45. It is of size 5 cm x 4 cm. it is pale pink in colour, smooth in appearance, firm in consistency and tender on palpation. No regional lymphadenopathy was observed. (Fig a &b) On Radiograph examination, a well-defined radiolucency with focal areas of calcifications in panoramic radiograph; along with well-defined hypodense lesion with expansion of buccal cortical plate extending from 35 crossing the midline till 46 involving impacted 43 was evident in Computed tomogram (Fig c & d). On aspiration a straw coloured fluid was obtained. Hence a provisional diagnosis of unicystic ameloblastoma/dentigerous cyst/odontogenic keratocyst was made and after incisional biopsy a histopathological diagnosis of adenomatoid odontogenic tumour was made. The lesion was excised in to under local anesthesia (Fig e) and a definitive diagnosis of cystic adenomatoid odontogenic tumour was done.

Clinical findings

Figure a:

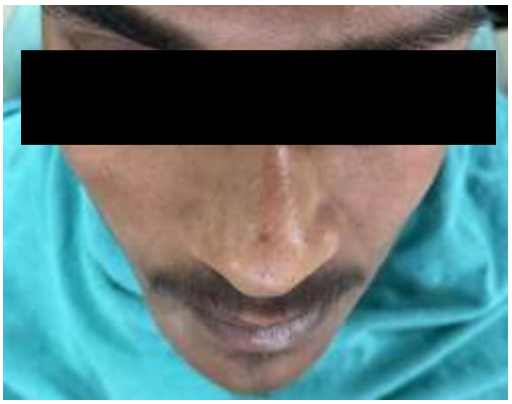
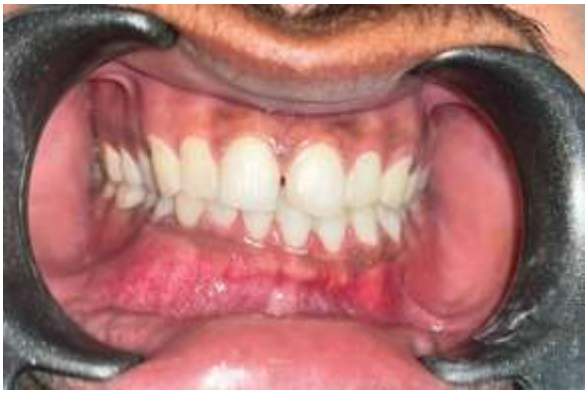


Figure b:



Well-defined hard unilateral swelling extending antero-posteriorly involving the gingiva from 83 to 45 . It is of size 5 cm x 4 cm

Radiological findings

Figure c: Well defined radiolucency in OPG extending from 35 crossing the midline till 46 involving impacted 43 with focal areas of calcifications

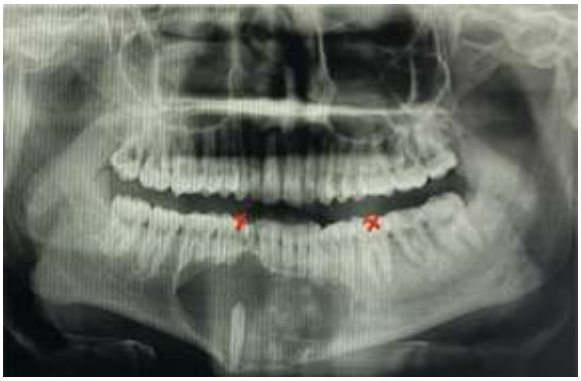
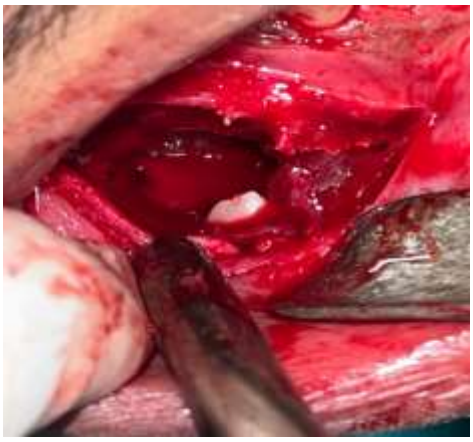


Figure d: well-defined hypodense lesion with expansion of buccal cortical plate extending from 35 crossing the midline till 46 involving impacted 43 was evident in Computed tomogram

Figure e: Intra-operatively impacted 43 with evident cystic cavity



Histological findings

Microscopically, Submitted H & E stained sections shows presence of odontogenic epithelial cells proliferating in numerous ducts within a scanty stroma. Duct like structure exhibits a central space filled with eosinophilic coagulum and lined by peripheral cuboidal cells showing hyperchromatic nuclei with scant cytoplasm and outer layer of loose stellate reticulum like cells. Numerous nests and rosettes like arrangements of odontogenic cells separated by thick fibro collagenous stroma are also evident. Small foci of calcification are seen within the stroma .Varying amount of dentinoid or amyloid like eosinophilic substance is also seen dispersed within the tumor. Spicules of bone is also evident along with blood vessels. (Fig f & g)

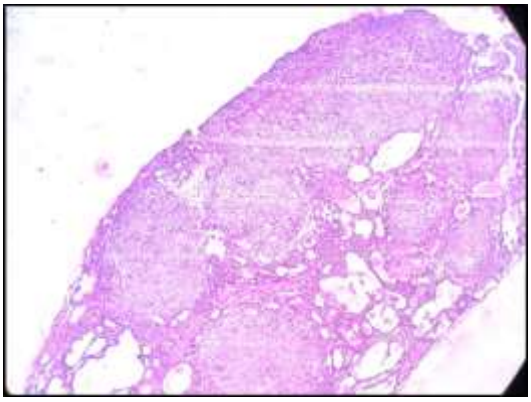


Figure f: Scanner View

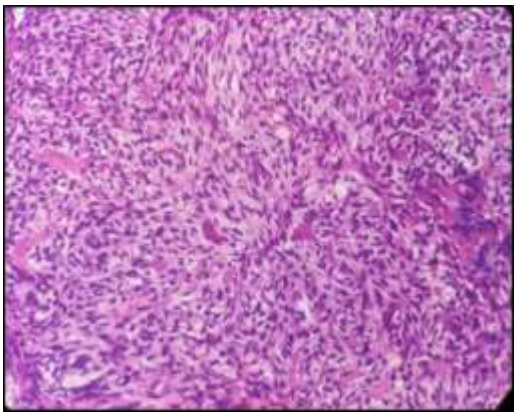


Figure g: 10x

Fig f and g: Excisional Biopsy: Odontogenic epithelial cells proliferating in numerous ducts filled with eosinophilic coagulum**.**

Discussion

The Adenomatoid Odontogenic Tumor (AOT) is a benign lesion typically found in the anterior maxillary bones, with a higher incidence among females in their second decade of life. It can manifest in intraosseous and peripheral forms. Radiographically, intrabony variants include follicular and extra-follicular types. The follicular type often presents as a well-defined, unilocular radiolucency associated with the crown and sometimes the root of an unerupted tooth, resembling a dentigerous cyst. However, a distinguishing feature is the association of the radiolucency with the root, which is typical in AOT but not in dentigerous cysts.

Histopathologically, AOT exhibits diverse patterns, leading to debates about its nature. Initially considered a hamartoma due to its limited size and lack of recurrence, some authors believe it's more akin to a benign neoplasm. The contention lies in whether it originates from the tooth follicle or the remnants of the dental lamina. Some propose the term "Adenomatoid Odontogenic Cyst (AOC)," suggesting it arises from the remnants of Hertwig's epithelial root sheath (HERS). Others argue for its origin from the dental lamina or its remnants.

Recent evidence suggests AOT may originate from the cystic lining, particularly in cases associated with dentigerous cysts. Immunohistochemical studies reveal similarities between the epithelium of reduced enamel epithelium (REE) and AOT, supporting the notion of REE as a progenitor of AOT. Additionally, cases where the tooth is entirely within the lumen may align with AOT arising from HERS. This perspective suggests that all AOTs may initially begin as cysts derived from either REE or HERS, with classical AOTs filling the cystic lumen entirely, while cystic variants show incomplete proliferation.

In summary, AOT exhibits diverse histopathological patterns, with debates about its origin ranging from the dental lamina to HERS. Evidence suggests it may originate from the cystic lining, particularly REE, with cases supporting its association with dentigerous cysts. This perspective offers insights into the pathogenesis of AOT and its variants.

Conclusion

Adenomatoid Odontogenic Tumor (AOT) is an uncommon, slow-growing tumor characterized by its painless and noninvasive nature, often leading to misdiagnosis as an odontogenic cyst. While typically affecting young individuals, predominantly females, and commonly localized in the anterior maxilla with an association with impacted canines. Notably, in our present case it exhibited distinct clinical and radiographic features not typically seen in conventional AOT presentations. In our findings, intraoral periapical radiographs proved most effective in highlighting radiopacities within AOT, presenting as discrete foci with a flocculent pattern amidst radiolucency, even with minimal calcified deposits. It is crucial to underscore that while AOT is rare, meticulous diagnosis and thorough interpretation of clinical and radiographic

findings can significantly aid in reaching an accurate diagnosis.

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