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Case Report

Unveiling the uncommon: Adenomatoid odontogenic tumor linked to an impacted mandibular canine

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Abstract

Adenomatoid odontogenic tumor (AOT) is an uncommon tumor that arises from the epithelial cells, accounting for only 3-7 % of all odontogenic tumors. It is a harmless, painless, non-invasive, and slow-growing benign lesion, with a frequency of and frequently mistaken for an odontogenic cyst during clinical examination. AOT primarily affects young people, with a higher occurrence in females, and commonly occurs around the crown of unerupted teeth in the second decade of life. This lesion is typically found in the maxillary jaw and is quite rare in the mandibular jaw. It is commonly linked to an impacted canine. AOT often resembles lesions similar to dentigerous cyst or ameloblastoma. AOT has three different types, follicular, extrafollicular, and peripheral. Follicular type is associated with the crown of an unerupted or impacted tooth, and the extrafollicular type, not associated with a tooth. Approximately 73% of central lesions are the follicular type.

Keyword: Admantinoma, Adenomatoid, Dentinoid, Amyloid, Liesegang rings.

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1. Introduction

Adenomatoid odontogenic tumor (AOT) is rare tumor that arises from the epithelial cells, accounting for only 3-7 % of all odontogenic tumors.[1] In 1905, Steensland first described a lesion that is now recognized as AOT.[1] Later, in 1948, Stafne referred to similar tumor as an “adamantinoma”. In 1953, Dreyblatt described it as an “adenomatoid odontogenic tumor.” highlighting its gland-like (adenomatoid) structures.1 Philipsen and Birn (1969) further refined its understanding, introducing the term Adenomatoid Odontogenic Tumor (AOT) and distinguishing it from other odontogenic tumors. [1-2] In 1971 World Health Organization (WHO) officially recognized AOT as a distinct benign epithelial odontogenic tumor. Max and Stern, in 2003, coined the name ‘adenomatoid odontogenic cyst’.[1-2] The terms like adamantinoma, adenoameloblastoma, ameloblastic adenomatoid tumor, epithelioma adamantinum, and teratomatous odontoma were used before term AOT being used. [1-2]

Adenomatoid odontogenic tumor (AOT) is uncommon, benign, slow growing, non-aggressive odontogenic tumor derived from odontogenic epithelium.[1-2] Cells of AOT originated from enamel organ epithelium. [1-2] It contains connective tissue elements and sometimes calcification as dentin or enamel-like structure.[1-2] On clinical examination AOT is often misdiagnosed as odontogenic cyst. It occurs in a wide range of 5 to 50 years, develop in second decade of life, with an average age of 16 years. AOT has 2:1 female predilection with two-third cases is associated with an unerupted tooth, and two-third affected teeth are impacted canines.[1-2]

2. A Case Report

A 13-year-old male patient visited to the Department of Oral Medicine and Radiology with a chief complaint of swelling in lower front region of jaw since 6 month. Initially the swelling was small, painless, localized and gradually increased to present size. There was no anesthesia or paresthesia of the lower lip, chin, or jaw, and there was no history of trauma.

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There was no h/o any pus discharge. Swelling became painful since 1 month. Then he visited to local hospital from where he got referred to this hospital. Now he reported to the hospital with present complaint of persistent pain and swelling.

Figure 1: Extraoral photograpgs

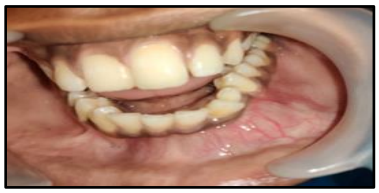

Figure 2: Intra Oral

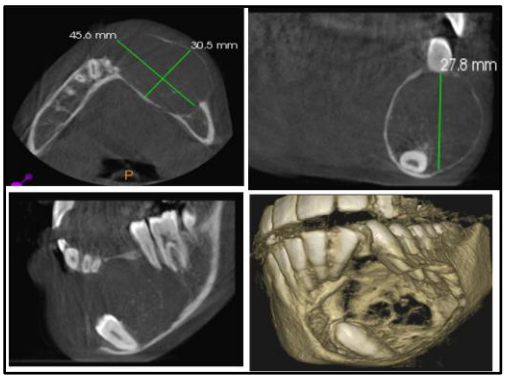

Figure 3: CBCT Photographs

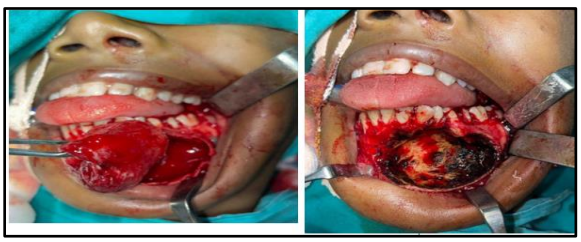

Figure 4: Post-operative photographs

Figure 5; Excised specimen

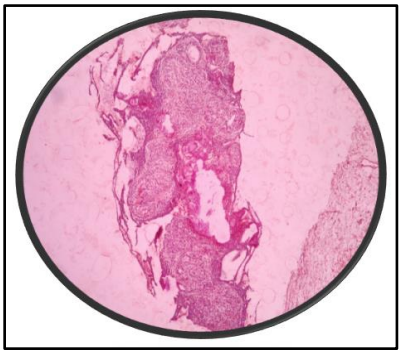

Figure 6: Histopathology slide

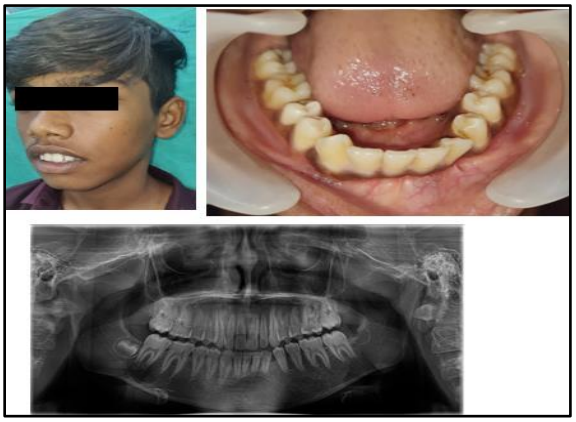

Figure 7: Follow Up Phototgraphs

3. Extra Oral Examination-(Figure 1,Figure 2,Figure 3)

3.1. On inspection

Facial asymmetry was present with a single, large, well defined, localized swelling noted at left parasymphysis region of mandible. The swelling extending anteroposteriorly from midline to 1 cm lateral to left corner of mouth and supero inferiorly from vermilion border of lip to inferior border of mandible. The size of swelling approximately AP x SI is 4-4.5 cm x 3.5-4 cm. No ulceration or pus discharge present.

3.2. On palpation

The swelling appears afebrile, non-tender, bony hard and fixed

4. Intra Oral examination

*4.1. Soft tissue examination: (**Figure 4**)*

Maxillary jaw was normal while mandibular jaw shows a well-defined intraoral swelling of approx. 4 x 3.7 cm present on the left side of lower buccal vestibule. Antero-posteriorly, swelling extends from distal surface of 42 to distal surface of 34. Supero-inferiorly, swelling present near to attached gingiva of 42,41,31,32,73,34 to the buccalvestibule. Medio-laterally, swelling has obliterated buccal vestibule opposite to 42, 41, 31, 32,73,34. Expansion of buccal and lingual cortices present. Surface of the swelling is slightly blanched, white in

colour compared to the adjacent mucosa. No ulceration or sinus tract present over the surface of the swelling. On palpation the swelling was non-tender, non-compressible, bony hard in consistency.

Hard tissue examination:

All permanent teeth were present with

1. Missing teeth 33
2. Retained deciduous teeth – 73
3. Mesial tipping of 31, 32
4. Mobility-

a) Grade I mobility wrt -31,32,41,42
b) Grade II mobility wrt - 34,35
c) Grade III mobility wrt – 73

5. Radiographic Investigation

CBCT Findings – **(Figure 5,Figure 6,**

Figure 7).

Location -A well-defined radiolucency of approx. 45.6x30.5x27.8 mm seen on the left body of mandible at the periapical region of 41,31,32,73,34,35,36. The radiolucency is found attached to obliquely placed impacted 33 at the inferior border of mandible. Antero-posteriorly the radiolucency is extending from mesial surface of 42 to mesial root of 36. Supero-inferiorly the radiolucency is extending from the alveolar crest to lower borderof mandible. Medio-laterally the radiolucency showed the expansion of buccal and lingual cortical plates adjacent to of 41,31,32,73,34,35,36.

6. Periphery

The periphery of the lesion is well defined, radiopaque, thin corticated margins surrounding the lesion.

7. Internal Structure

The internal structure of the lesion is multilocular with thin wispy septae divides the lesion. The multilocular appearance is similar to soap bubbles. The septae originates at the right angle to the periphery.

8. Effect on the Surrounding Structures

Diffuse osteopenia present. Thinning and expansion of buccal cortical plate with 41,31,32,73,34,35,36 region. Thinning and erosion of lingual cortical plate with 32,73,34,35,36 region. Inferior alveolar nerve canal displaced inferiorly.

9. Effect on Adjacent Teeth

There is localized complete loss of lamina dura seen with 32,73,34,35. There is thinning and partial disruption of lamina dura seen with 31,36. Root resorption and blunting of root apex of 31,32,83,34,35.

A radiographic diagnosis of adenomatoid odontogenic tumor (AOT) was given by considering the multiple scattered radiopaque flecks in the lesion associated with an unerupted impacted canine 33 and a soft tissue capsule. But a multilocular appearance with large size of an AOT is unusual. Hence, a differential diagnosis of other multilocular mixed lesions such as calcifying odontogenic cyst and calcifying epithelial odontogenic tumor was also considered.

The lesion was surgically enucleated with removal of tumor mass, impacted canine and over retained deciduous tooth 73. The tumor mass was then send for histopathological investigation. Patient was kept on regular follow up.

10. Histopathological Examination

1. The H/E stained section of single soft tissue bit exhibits cystic lining with non-keratinized odontogenic epithelium which is 2-3 cells layers thin, proliferating in the form of solid nests, whorled nodules, rosettes, ductal and stream pattern, cuboidal to low columnar cells with hyperchromatic polarized nuclei form single layer duct- like structures and double layered rosettes.
2. Few duct-like structures show an eosinophilic rim suggestive of dentinoid or amyloid like material at the periphery of the lumen.
3. Focal area of tumor cells is spindle and polygonal shaped with sparse connective tissue.
4. Anastomosing strands of basaloid cells are present in plexiform pattern focally and cribriform pattern at the periphery.
5. Focal areas of calcification are also seen. Thick fibrous capsules is evident at the periphery with parallelly arranged collagen fibers.
6. Overall features suggestive of “Adenomatoid odontogenic tumor”

So, summarizing all the investigation, the final diagnosis was given as Adenomatoid odontogenic tumor associated with impacted 33. After eight months of follow-up, clinical examination revealed complete resolution of the swelling. Follow up OPG shows complete healing and new bone formation.

11. Discussion

Adenomatoid odontogenic tumor (AOT) is an uncommon benign epithelial lesion of odontogenic origin, comprising approximately 3–7% of all odontogenic tumors.[1-2] It was historically misclassified due to its varied histological presentation, but the World Health Organization (WHO) recognized it as a distinct entity in 1971[3] AOT is often referred to as the “two-thirds tumor” due to its occurrence in females (2/3 of cases), its predilection for the maxilla (2/3 of cases), and its association with unerupted canines (2/3 of cases).[4]

The presented case is atypical in multiple respects. Firstly, it was located in the mandible, a less common site for AOT. The maxilla is involved in 65–75% of cases, with the anterior region being the most frequent location.[5] Mandibular

involvement, particularly in the parasymphysis region as seen here, is rare, representing only about 13–25% of AOT cases.[6]

Secondly, the lesion was associated with an impacted mandibular canine, which is unusual, given the lesion's known predilection for the maxillary canine. A study by Philipsen and Reichart found that only a minority of follicular AOTs involve the mandibular canine.[7] The radiographic appearance of this case also presents an atypical multilocular radiolucency, whereas classic AOTs are usually unilocular and may contain fine calcifications or snowflake-like radiopacities.[8]

The clinical presentation in this case—a slowly enlarging, initially painless swelling in a young adolescent—is consistent with the behavior of AOT. Typically, these tumors are asymptomatic, slow-growing, and often discovered incidentally or during the investigation of delayed tooth eruption.[9] However, the presence of pain in this patient during the later stages of growth is somewhat atypical, though it may be attributed to secondary infection or cortical expansion with nerve impingement, which is rare in AOT.[10]

Radiographically, AOT may mimic other odontogenic cysts and tumors. In this case, the initial differential diagnosis included calcifying epithelial odontogenic tumor (CEOT), and calcifying odontogenic cyst (COC). The multilocular appearance and significant size deviate from classical AOT radiology. Although most AOTs are unilocular, multilocular radiolucencies have occasionally been reported, particularly in larger lesions.[11] The presence of radiopaque flecks, however, along with a well-demarcated corticated border and association with an unerupted tooth, strongly favored AOT.[12]

Histopathologically, the hallmark of AOT is the presence of duct-like structures, rosette-like formations, spindle-shaped epithelial cells, and focal calcifications, all of which were present in this case.[13] The presence of eosinophilic material resembles amyloid or dentinoid, and basaloid cells arranged in plexiform and cribriform patterns, further confirmed the diagnosis. These features help distinguish AOT from ameloblastoma, which lacks duct-like structures, and from CEOT, which often shows Liesegang rings and prominent amyloid deposits.[14]

The treatment of choice for AOT is conservative surgical enucleation, with removal of the associated impacted tooth if present. Due to the tumor's benign and encapsulated nature, recurrence is extremely rare[15] In this case, complete enucleation along with extraction of the impacted mandibular canine was performed successfully, with no signs of recurrence at follow-up.

This case contributes to the limited pool of mandibular AOTs associated with impacted canines and underscores the importance of considering AOT in the differential diagnosis of mixed radiolucent lesions in young individuals, even in uncommon locations. Accurate diagnosis through clinical, radiological, and histopathological correlation is essential to avoid overtreatment or misdiagnosis.

12. Conclusion

Adenomatoid odontogenic tumor (AOT) is a rare, benign lesion typically seen in the anterior maxilla and associated with unerupted teeth, most commonly maxillary canines. This case highlights an unusual presentation of AOT in the mandibular parasymphysis region, associated with an impacted mandibular canine and displaying a multilocular radiographic appearance.[15] Despite its rare site and atypical imaging features, the lesion was successfully diagnosed through comprehensive clinical, radiographic, and histopathological analysis. Timely surgical enucleation led to favorable outcomes without recurrence. This report emphasizes the need for clinicians to include AOT in the differential diagnosis of jaw swellings in adolescents, especially when associated with impacted teeth, regardless of location.

13. Source of Funding

None.

14. Conflict of Interest

None.

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