ADENOMATOID ODONTOGENIC TUMOR WITH RARE CLINICAL AND RADIOLOGICAL PRESENTATION- A CASE REPORT

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ABSTRACT:
The Adenomatoid odontogenic tumor (AOT) is a benign (hamartomatous) noninvasive lesion but progressive growth, constituting only 3% of all odontogenic tumors. Most common site of AOT is maxillary anterior region especially canine region. Most common variety of AOT is Follicular variety (73%) which is associated with impacted tooth (maxillary canine). Females are most commonly affected than males. We report on a rare case of follicular AOT in the mandibular anterior region seen in an 18 years old male patient. Diagnosis of adenomatoid odontogenic tumor should be considered when the clinician is presented with a corticated radiolucency in the anterior lower jaw, especially in teens and young adults.

Key words: Adenomatoid odontogenic tumor; Maxilla; Root resorption;

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INTRODUCTION:
The odontogenic tumors are a diverse group of lesions that represent the deviation from normal odontogenesis. The AOT is an epithelial tumor with an inductive effect on odontogenic ectomesenchyme. AOT can be clearly distinguishable from the classic intraosseous, infiltrative ameloblastoma. It was suggested to abandon the previously used term adenameloblastoma; Philipsen and Brin [1] introduced the above term (AOT), which was adopted by the WHO classification in 1971 [2]. The benign (hamartomatous) noninvasive AOT appears in 3 clinic topographic variants; 1)
Follicular, 2) Extra follicular and 3) peripheral.

Follicular and extra follicular variants are both intra-bony or central tumors and account for 97% of all AOTs of which 73% are of the follicular type [3]. The extra follicular variant is not associated with an unerupted tooth like the follicular variant, and the well defined, unilocular radiolucency is found between, above, or superimposed on the roots of erupted teeth. It is characteristic that the rare sub variant mimicking a periapical lesion is in fact located palatally (or lingually) to the tooth involved [4]. Sixty – nine percent of AOTs are diagnosed in the second decade of life, and more than half of the cases (53%) occur during the teenage years (13 to 19 years of age). The females to male ratio for all age groups and AOT variants together is very close to 2:1 and predominantly seen in the maxilla (maxilla : mandible = 2.6:1) [5].

CASE REPORT:

An 18 year old male reported with a swelling of the right lower jaw region since 2 months. The swelling gradually progressed to attain its present size in two months duration. Extraorally (Figure 1) swelling measured 4x5cm and extended mesio-distally from left parasymphisis to right parasymphisis region. Supero-inferiorly extended from 1.0cm below the vermilion border of the lower lip to the sub mental region; it covered the whole chin region with diffuse margins. Intraorally (Figures 2 & 3) the swelling extended labially from distal aspect of region 32 to mesial aspect of region 44, with vestibular obliteration. Lingually a diffuse swelling was noted extended from distal aspect of region 41 to mesial aspect of region 44. The consistency was firm. Grade 3 mobility i.r.t over retained 83 (lower right deciduous canine). 31, 41, 42 showed grade 2 mobility.

We came to the Provisional diagnosis of dentigerous cyst and differential diagnosis of calcifying epithelial odontogenic cyst, central giant cell granuloma, adenomatoid odontogenic tumor was considered.

INVESTIGATIONS:

Radiographically Intra oral periapical radiograph (IOPA) shows well defined unilocular radiolucency measuring about 4x3 cm extending mesiodistally from distal aspect of 45 region to other side of the radiolucency can’t be make out with an impacted dilacerated 43, radiolucency extending at the apex of the root. supero- inferiorly extending from alveolar crest region i.r.t 41,42, over retained deciduous right canine (83) to lower border of the mandible causing thinning of the lower border of the mandible. External root resorption i.r.t 83 (right lower deciduous canine) with displacement of teeth roots i.r.t 41,42, 44 & 45.
Occlusal radiograph shows well defined buccolingual cortical expansion (Hydraulic expansion), extending from distal aspect of 33 region to mesial aspect of 46. (Figure 5)

Orthopantomogram (OPG) shows well defined radiolucency measuring about 3x4cm extending mesio-distally from region 33 to 45, supero-inferiorly from alveolar crest region from 42, 44 region to lower border of the mandible causing thinning and expansion, which is surrounded by well-defined corticated radiolucency. Dilacerated apically displaced impacted tooth is embedded within the radiolucency which is, extending till apex of the 43. Loss of lamina dura i.r.t apical 1/3rd of 31, 32, 41 to 45 regions (Figure 6).

Pulpal vitality test was done, except 83 all the teeth showed positive response.

FNAC (Fine needle aspiration cytology) was performed showed straw – color aspiration, subjected for protein investigation (3.9mg%), other microscopic examination like cholesterol crystal examination, which was negative (Figure 7).

Surgical enucleation of the tumor for histopathological examination was performed under local anesthesia.

Microscopically tissue section of the specimen showed odontogenic epithelium proliferating in the form of whorls with rosset like pattern, showing globular calcifications, duct like areas are seen with hyperchromatic epithelial cells suggestive of adenomatoid odontogenic tumor (Figures 8 A & B)

DISCUSSION:

AOT is an uncommon tumor or benign, hamartomatous growth derived from odontogenic epithelium. These tumors tend to develop in younger people and are more common among women, but in our case it was found in adult male patient [3].

Three clinico-pathological variants are well recognized; follicular, extra follicular, and peripheral. The follicular and extra follicular variants are intra-osseous and account for about 96%. The maxilla, often together with an unerupted canine, is most commonly affected than the mandible. In our case it was found in mandible with an unerupted canine seen in only 35.7% of cases [3, 5].

Our case we found separate follicular AOT associated with a right mandibular canine, in a 18 year old boy with radiographic findings mimicking dentigerous cyst. Radiographically unilocular radiolucency with no internal calcification has been seen in only 27% of cases. To differentiate from dentigerous cyst,
CEOC, Radicular cyst, and CGCG, mandibular premolar-molar region with an impacted tooth is the common site for dentigerous cyst; maxillary canine region is the most common site for AOT.

Dentigerous cyst radio graphically appears as the radiolucency attached to the cementoenamel junction; in case of AOT the radiolucency extends more apically beyond Cemento enamel junction as seen in our case. CEOC (Calcifying epithelial odontogenic cyst) is most commonly seen in either maxilla or mandibular region anterior to first molar of young adults, not associated with an impacted tooth, radiographically unilocular radiolucency with internal calcification are seen. Radicular cyst also can be considered in the differential diagnosis because, pulpal vitality test shows no response i.r.t 83, and the periodontal ligament and lamina dura were not found to be intact around involved teeth as seen in our case. Radiographically radicular cyst presents as a well defined unilocular radiolucent area, thin rim of cortical bone, larger than 1.5cm, with displacement of adjacent teeth. In our case shows buccolingual cortical expansion with well defined unilocular radiolucency approximately about 4cm with displacement of adjacent teeth. Most common site for radicular cyst is maxillary anterior region but in our case it was found in mandibular anterior region and on aspiration it yields yellow color pus discharge, but in our case it yielded straw color fluid.

Central giant cell granuloma (CGCG) is an aggressive lesion commonly seen in females younger than 30 years; mandibular anterior region is the common site, crossing midline. Radiographically it shows unilocular or multilocular radiolucency with wispy septae, displacement of teeth and root resorption are evident [6].

AOT is usually well – encapsulated, so conservative enucleation and currettage is the most common treatment modality for this tumor, recurrence is extremely rare. Care full follow-up examinations should be conducted in the case [3, 8, 10].

**CONCLUSION:**

It should be emphasized that careful diagnostic procedures and adequate interpretation of clinical and radiographic findings and differential diagnosis can be listed out to arrive at a correct diagnosis as in our case the AOT is mimicking dentigerous cyst. The final diagnosis of an AOT was arrived by histologic examination.
Unusual findings seen in our case related to typical features of an adenomatoid odontogenic tumor (AOT).

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<tr>
<th>Typical features of most AOT</th>
<th>Unusual findings in our case</th>
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<tr>
<td>Most commonly seen in females, maxillary canine being the most common site [5].</td>
<td>Seen in male patient with manibular (35%) Canine region.</td>
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<td>Cortical plate penetration rare size usually does not exceed 1-3 cm [3,7].</td>
<td>Cortical expansion is seen with AOT Size exceeded more than 4 cms.</td>
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<td>Unilocular radiolucent area with radiopaque specks Associated with impacted teeth [4].</td>
<td>Unilocular radiolucent area without radiopaque specks (seen in only 27% of cases).</td>
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<tr>
<td>Root resorption extremely rare; only four cases Reported to date to our knowledge [7 – 10].</td>
<td>Root resorption of the over-retained deciduous canine, with displacement of adjacent teeth (premolars).</td>
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Figure 1: Extra oral photograph of the patient

Figure 2: Intra-oral photograph showing lingual swelling in the anterior region of the mandible
Figure 3: Intra-oral photograph showing lower labial vestibular obliteration

Figure 4: Patient Intra-oral Periapical radiograph showing well defined radiolucency with impacted dilacerated canine. Also notice displaced premolars

Figure 5: Mandibular occlusal radiograph showing buccal and lingual cortical expansion with no cortical perforation
Figure 6: Orthopantomogram showing well defined radiolucency with impacted canine and displaced premolars, overretained deciduous canine

Figure 7: Fine needle aspiration cytology showing straw coloured aspirate

Figure 8: Pictomicrograph 40X magnification (A) and 100X magnification (B) (H & E). Showing odontogenic epithelium proliferating in the form of whorls with rosset like pattern, showing globular
calcifications, duct like areas are seen with hyperchromatic epithelial cells suggestive of adenomatoid odontogenic tumor

REFERENCES:


5. Philipsen HP, Reichart PA, Nikai H. The adenomatoid odontogenic tumor (AOT); An update. Oral med pathol 1997;2;55-60.

6. Stuart C. White, Michael J. Pharoah; Oral radiology; 6th edition; Benign Tumors of the jaws; Chapter 22; 383-385.


