CASE REPORT

An Unusual Site of Adenomatoid Odontogenic Tumor Presenting as Periapical (Radicular) Cyst: A Rare Case Report

Anand Kumar, Jagat Reddy, Siddharth Gupta, Namita Raghav, MunBhawni Bagga

1Professor and Head, Department of Oral Medicine Diagnosis and Radiology, KD Dental College and Hospital Mathura, Uttar Pradesh, India
2Reader, Department of Oral Medicine Diagnosis and Radiology, KD Dental College and Hospital, Mathura, Uttar Pradesh, India
3,4Senior Lecturer, Department of Oral Medicine Diagnosis and Radiology, KD Dental College and Hospital, Mathura, Uttar Pradesh, India
5Postgraduate Student, Department of Oral Medicine Diagnosis and Radiology, KD Dental College and Hospital, Mathura, Uttar Pradesh, India

Correspondence: C Anand Kumar, Professor and Head, Department of Oral Medicine Diagnosis and Radiology, KD Dental College and Hospital, Mathura, Uttar Pradesh, India, e-mail: anandmds@rediffmail.com

Abstract
The adenomatoid odontogenic tumor is a rare odontogenic tumor often misdiagnosed as an odontogenic cyst, constituting only 3% of all odontogenic tumors. Though odontogenic in origin but it appeared as duct like structures, often interspersed throughout the lesion which gives a glandular, i.e. Adenomatoid appearance.

Here we are presenting a rare case report of an unusual site of extrafollicular adenomatoid odontogenic tumor in the mandible w.r.t 32, 33, 34 and 35 mimicking periapical disease clinical and radiographically. However, 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.

Keywords: Adamantinoma, extrafollicular, odontogenic tumor, unilocular radiolucency and surgical enucleation.

INTRODUCTION
The odontogenic cyst and tumors are a diverse group of lesions that represent the deviation from normal odontogenesis. The adenomatoid odontogenic tumor (AOT) is an epithelial tumor with an inductive effect on odontogenic ectomesenchyme. The tumor is a benign (hamartomatous), noninvasive lesion with slow but progressive growth. It constitutes an entity within the odontogenic tumors, clearly distinguishable from the classic intraosseous, infiltrative ameloblastoma. It was first described by Driebladt in 1907 as a pseudoadenoma adantaminoma.2

AOT accounts for 2.2 to 7.5% of all odontogenic tumors. Philipsen and Birn proposed the name adenomatoid odontogenic tumor in 1969.3 In 1971, the name was adopted by WHO in their histological typing for odontogenic cyst, tumors, jaw cysts and allied lesions. In a series of publications, it has been clarified that the mean age of the uncommon distinct odontogenic neoplasm is usually 13.2 years and female male ratio being 2.3:1 and predominantly seen in the maxilla (maxilla : mandible = 2.6:1).5,4

There are three variants of AOT:
1. Follicular
2. Extrafollicular and
3. Peripheral (extraosseous).

Follicular and extrafollicular variants account for 97% of all AOTs of which 73% are of the follicular type.5 The extrafollicular variant radiographically present as a unilocular radiolucency found between, above, or superimposed on the roots of erupted teeth. It is characteristic that the rare subvariant mimicking a periapical lesion is, in fact, located palatally (or lingually) to the “involved” tooth. It has been theorized that the complex system of the dental lamina or its remnants is the likely origin of the AOT mimicking a periapical cyst.6,7

CASE REPORT
A 17-year-old female patient reported to the department of Oral Medicine and Radiology, with a chief complaint of swelling in left lower jaw region since 3 months. Beside an associated history of trauma since three years, the swelling gradually progressed to attain its present size in three months duration with no history of pain, discharge and numbness revealed. On extraoral examination, a solitary ovoid swelling was present on left lower 1/3 of face extending superiorly from left lower lip to inferiorly 1 cm above inferior border of mandible. Anteriorly the swelling extends 0.5 cm away from midline to middle of the body of mandible; measuring about 1.5 × 1.5 cm in size, with smooth surface and diffuse margins. It was not tender, firm and fixed to underlying bony structure on palpation. Regional lymph nodes were not palpable.

On intraoral examination, a solitary ovoid swelling was present on buccal and lingual vestibule of left lower lateral incisor, canine and premolar region extending from distal aspect of 32 to distal aspect of 35. Swelling was lobulated with clearly defined margins, and overlying mucosa was intact with no ulceration and sinus discharge (Fig. 1).

Abstract

The adenomatoid odontogenic tumor is a rare odontogenic tumor often misdiagnosed as an odontogenic cyst, constituting only 3% of all odontogenic tumors. Though odontogenic in origin but it appeared as duct like structures, often interspersed throughout the lesion which gives a glandular, i.e. Adenomatoid appearance.

Here we are presenting a rare case report of an unusual site of extrafollicular adenomatoid odontogenic tumor in the mandible w.r.t 32, 33, 34 and 35 mimicking periapical disease clinical and radiographically. However, 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.

Keywords: Adamantinoma, extrafollicular, odontogenic tumor, unilocular radiolucency and surgical enucleation.

INTRODUCTION
The odontogenic cyst and tumors are a diverse group of lesions that represent the deviation from normal odontogenesis. The adenomatoid odontogenic tumor (AOT) is an epithelial tumor with an inductive effect on odontogenic ectomesenchyme. The tumor is a benign (hamartomatous), noninvasive lesion with slow but progressive growth. It constitutes an entity within the odontogenic tumors, clearly distinguishable from the classic intraosseous, infiltrative ameloblastoma. It was first described by Driebladt in 1907 as a pseudoadenoma adantaminoma.2

AOT accounts for 2.2 to 7.5% of all odontogenic tumors. Philipsen and Birn proposed the name adenomatoid odontogenic tumor in 1969.3 In 1971, the name was adopted by WHO in their histological typing for odontogenic cyst, tumors, jaw cysts and allied lesions. In a series of publications, it has been clarified that the mean age of the uncommon distinct odontogenic neoplasm is usually 13.2 years and female male ratio being 2.3:1 and predominantly seen in the maxilla (maxilla : mandible = 2.6:1).5,4

There are three variants of AOT:
1. Follicular
2. Extrafollicular and
3. Peripheral (extraosseous).

Follicular and extrafollicular variants account for 97% of all AOTs of which 73% are of the follicular type.5 The extrafollicular variant radiographically present as a unilocular radiolucency found between, above, or superimposed on the roots of erupted teeth. It is characteristic that the rare subvariant mimicking a periapical lesion is, in fact, located palatally (or lingually) to the “involved” tooth. It has been theorized that the complex system of the dental lamina or its remnants is the likely origin of the AOT mimicking a periapical cyst.6,7

CASE REPORT
A 17-year-old female patient reported to the department of Oral Medicine and Radiology, with a chief complaint of swelling in left lower jaw region since 3 months. Beside an associated history of trauma since three years, the swelling gradually progressed to attain its present size in three months duration with no history of pain, discharge and numbness revealed. On extraoral examination, a solitary ovoid swelling was present on left lower 1/3 of face extending superiorly from left lower lip to inferiorly 1 cm above inferior border of mandible. Anteriorly the swelling extends 0.5 cm away from midline to middle of the body of mandible; measuring about 1.5 × 1.5 cm in size, with smooth surface and diffuse margins. It was not tender, firm and fixed to underlying bony structure on palpation. Regional lymph nodes were not palpable.

On intraoral examination, a solitary ovoid swelling was present on buccal and lingual vestibule of left lower lateral incisor, canine and premolar region extending from distal aspect of 32 to distal aspect of 35. Swelling was lobulated with clearly defined margins, and overlying mucosa was intact with no ulceration and sinus discharge (Fig. 1).
On palpation, a solitary ovoid swelling which was firm in consistency showed no evidence of discharge on digital pressure. It was not tender with no pulsations evident. Egg shell crackling was present on palpation. The associated teeth were found to be mobile and nonvital.

Intraoral periapical (IOPA) radiograph w.r.t 32, 33, 34 and 35; mandibular cross-sectional occlusal radiograph and orthopantomograph were carried out. It revealed well-defined periapical radiolucency measuring about 2 × 1.5 cm with fine calcifications surrounded by sclerotic border extending from distal of 31 to distal of 35. Teeth 33 and 34 were displaced and revealed root resorption (Figs 2 and 3).

Fine needle aspiration was carried out and straw colored fluid was aspirated. Microscopic examination revealed histiocytes, mast cells; polymorph macrophages and few foam cells were present.

Various differential diagnosis such as periapical cyst, traumatic bone cyst, aneurysmal bone cyst, ameloblastoma were put forth.

Surgical enucleation of the tumor was carried out under local anesthesia and sample was send for histopathologic examination. Since the tumor extended below the roots of 32, 33, 34 and 35 these teeth were removed. The tumor cavity was removed to reduce the dead space, the cavity was packed with three large pieces of Gelfoam and the mucoperiosteal flap was replaced into contact with the packing. Healing was uneventful, with no evidence of recurrence after surgery. After 6 months, prosthetic rehabilitation of the patient was performed and implants were placed w.r.t residual alveolar ridge of 32, 33, 34 and 35.

Histopathological report showed that a cystic cavity lined by stratified squamous epithelium with multinodular proliferation of spindle, columnar and cuboidal cells arranged in form of sheets, strands and whorled masses. Epithelial cells revealed rosette like structure about a central space containing eosinophilic substance. The characteristic duct like structure with lumen lined by single layer of cuboidal and columnar cells with scattered eosinophilic substance and calcifications were also present, suggestive of extrafollicular adenomatoid odontogenic tumor w.r.t anterior portion of left side of mandible (Fig. 4).

DISCUSSION
The AOT comprises approximately 3% of all odontogenic tumors, ranking behind odontoma, periapical cemental dysplasia (cementoma), myxoma, and ameloblastoma. The origin of the
An Unusual Site of Adenomatoid Odontogenic Tumor Presenting as Periapical (Radicular) Cyst: A Rare Case Report

AOT is controversial. However, evidence also exists as in our case also that the tumor could be derived from epithelial remnants of the dental lamina complex system. The lesion then presents radiographically as a residual, developmental, lateral, periodontal, or radicular cyst, depending on the location of the epithelial cells of rest.

The case report illustrates characteristic clinical and radiographic features of the extrafollicular variant of the AOT mimicking a periapical lesion at an unusual site that is mandible. There were in fact, a number of clinicoradiologic indicators that could have suggested that this case in a 17-year-old girl was a possible extrafollicular AOT mimicking radicular cyst, although this subvariant is indeed a rarity. The age distribution of patients with AOT shows a peak in the second decade the female sex is almost twice as often affected as the male. The incisor/canine region is often the site of an extrafollicular AOT in which the tumor produces a slowly enlarging swelling along with the maxillary predilection of tumor. A distinct radiopaque border of the unilocular radiolucency is typical of the radiographic manifestation of an AOT. The periodontal ligament and lamina dura were not found to be intact around involved teeth an important finding that should make a periapical radiolucent lesion such as periapical cyst or granuloma more likely. The involved teeth were nonvital on pulp testing mimicking a radicular cyst. The other rare features in our case were history of trauma favors it to be more likely a radicular cyst and egg shell crackling mimicking it to be a unicystic ameloblastoma. Root resorption is seldom reported. The patient we described in this report also presented resorption of the lower left canine and first premolar. Conservative surgical enucleation or curettage is the treatment of choice with only rare recurrence.

The patient we described in this case report had no recurrence and regular follow-up was done after local excision.

CONCLUSION

It should be emphasized again that this particular subvariant is very rare indeed; being the least reported presentation of the extrafollicular variant. Only careful diagnostic procedures and adequate interpretation of clinical and radiographic findings may result in a correct diagnosis, which otherwise may result in unnecessary endodontic treatment. The final diagnosis of an AOT was arrived by histologic examination of the removed tissue mass.

REFERENCES