



Adenomatoid Odontogenic Tumour Mimicking Dentigerous Cyst Associated with Mandibular Lateral Incisor: An Unusal Case

Dr. Sheeraz Badal¹, Dr Rahul Laturiya², Dr Paras Doshi³, Dr Varsha Jaju³

¹HOD & Professor, ²Professor, ^{3,4}3rd year PG,

Department of Oral and Maxillofacial Surgery, MIDSR Dental College, Latur, Maharashtra.

Corresponding author: Dr Paras Doshi³

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ABSTRACT:

Adenomatoid odontogenic tumor (AOT) is a rare odontogenic tumor which is often misdiagnosed as odontogenic cyst. This clinical entity has been regarded as a benign odontogenic tumor, sharing its clinical and radiographic features with those of dentigerous cysts. It is a slow growing, non-invasive benign lesion of odontogenic epithelial origin. The maxilla is more afflicted than the mandible, and the anterior part of the jaw is more persistently involved than the posterior part.. An impacted maxillary canine is the most common tooth to be associated with an adenomatoid odontogenic tumor (AOT). Here is a case report of a 16 year old girl who presented with a well defined unilocular radiolucency involving an impacted mandibular lateral in the anterior mandible, which resembled a dentigerous cyst. Enucleation done and excised lesion was histopathologically confirmed to be an Adenomatoid odontogenic tumor (AOT)

Introduction

Adenomatoid odontogenic tumour (AOT), called as the master of disguise, is a relatively uncommon distinct odontogenic neoplasm that was first described by Steensland in 1905.^[1] It is slowly progressive growth that accounts for 2.2–13% of all odontogenic tumours. Philipsen and Birn proposed the name AOT, which was widely accepted and adopted by the World Health Organization classification of odontogenic tumors in 1971.^[2] AOT appears in three clinicopathologic variants: follicular, extrafollicular and peripheral. The follicular and extrafollicular variants are both intrabony, and account for approximately 96% of all AOT's of which 71% are of follicular type.^[2] The follicular type is associated with an unerupted tooth whereas extrafollicular type has no relation with an impacted tooth and the peripheral variant is attached to the gingival structures. All AOT variants demonstrate identical histology which indicates a common derivation. It is hypothesised that the lesion originates from reduced enamel epithelium, enamel organ epithelium and cell rests of Malassez.^[3,4] Recently, it has been suggested that the lesion may arise from remnants of the dental lamina associated with the gubernacular cord. It is also known as 'two thirds tumor,' because 2/3rd of cases occur in the maxilla, 2/3rd occur in young females, 2/3rd of the cases

are associated with un-erupted teeth, and 2/3rd of the affected teeth are canines.^[5] But this fascinating entity may camouflage as other odontogenic tumours/cysts, as seen in our three cases, two of which are atypical and one is a typical presentation of this tumour.

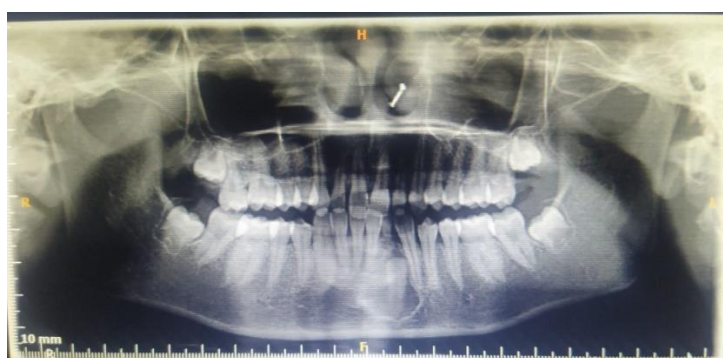
Case History

A 16-year-old female, patient presented with an incidental finding of a well-defined unilocular radiolucency was noticed involving an impacted tooth in the anterior region of the mandible on the panoramic view which was taken in lieu of orthodontic therapy. On clinical examination, there was no significant finding such as swelling, vestibular obliteration. There was no history of trauma. The patient gave a history of retained deciduous lateral incisor. There were no significant, palpable lymph nodes. There was no paresthesia of the lower lip. On radiographic examination, a well-defined unilocular radiolucency involving an impacted mandibular lateral was noticed. The lesion extended from the distal aspect of the central incisor to the mesial aspect of the canine anteroposteriorly on the left side and 1 cm away from the lower border of the mandible. Brick red-colored fluid was perceived on aspiration, which evokes cystic nature. Based on clinical and radiographic findings, it was provisionally diagnosed as a dentigerous cyst.



(a) (b)

(c)



(d)



(e)

image (a),(b),(c),(e) pre-operative extra oral and intra oral photos and

image (d) showing impacted lateral incisor along with well-defined unilocular radiolucency

The patient was posted for surgery and the lesion was enucleated under local anesthesia. On curetting it was hinge with a soft-tissue mass was fixed to the impacted lateral incisor. On enucleation, the soft-tissue mass and tooth came out completely. Soft-tissue was fixed on lateral aspect of the root, measuring 0.5×1 cm² in size. The specimen was sent for histopathological examination. Microscopically, the lesion was encapsulated by a thick connective tissue capsule. It showed sheets, ducts, and whorls of darkly staining ovoid

to round epithelial cells suggestive of odontogenic epithelial origin [Figure 4]. Duct-like structures were lined by columnar cells. A few basophilic calcifications were also observed. Small cystic areas containing degenerated cell debris were noted in focal areas. The supporting connective tissue stroma was loose and less cellular in nature. There was a typical thin layer of eosinophilic material at the periphery of the lumen. These features were suggestive of an AOT.



Image showing Stepwise procedure of enucleation

Dentigerous cyst, odontogenic keratocyst, unicystic ameloblastoma, and ameloblastic fibroma were considered in the list of differential diagnoses. A dentigerous cyst is always associated with an impacted tooth usually involves mandibular and maxillary posterior regions and maxillary canine areas. On radiography, it displayed a radiolucent area associated with an impacted tooth. An odontogenic keratocyst is a benign uni- or multicystic odontogenic lesion that shows aggressive and infiltrative behavior and commonly affects the posterior mandible. An odontogenic keratocyst can found in the anterior region of the mandible and can imitate a dentigerous cyst. A unicystic ameloblastoma shows clinical and radiological features like other cystic lesions of odontogenic origin, but it usually present in the posterior mandible. AOT shares its clinical and radiographic features with some of the lesions of odontogenic origin, but a final diagnosis is made only after the histopathological evaluation of the lesion

Discussion:

Adenomatoid odontogenic tumor is a benign, non-invasive lesion comprising of approximately 3% of all odontogenic tumors ranks after odontomes, cementoblastomas, myxomas and ameloblastomas AOT is a slow-growing lesion usually develops in anterior

maxilla and rarely noticed in the mandible, and it causes a painless expansion of the jaws. It tend to affect the deciduous tooth-bearing areas of maxilla and mandible. It has a predilection for female patients, with a female-to-male ratio of 1.9:1.^[1,5] Age range of 3-82 years, 90% diagnosed before the age of 30 years.^[2] AOT was first described as a distinct clinical entity by Stafne in 1948. Several authors have used various terms to describe AOT [Table 1]. The lesion may rarely exhibit aggressive behavior, such as becoming unusually large or spreading into the intracranial space.

AOT shows similar radiological findings with other odontogenic lesions, such as a dentigerous cyst, calcifying odontogenic cyst, ameloblastoma, or keratocystic odontogenic tumor. Corticated radiolucencies with small radiopaque foci in young adult patients should be considered in differential diagnosis of AOT.^[6] An AOT covers the crown and the root, whereas a dentigerous cyst cover only the crown attached to it. It was reported in prior studies that the intraosseous follicular variant of AOT showed a well-delineated, unilocular radiolucency. Multilocular, mixed radiolucent-radiopaque appearance with an impacted tooth, unusually large size, and aggressive behavior have also been recorded.^[7]



The origin of AOT is controversial, but as it arises in the tooth bearing area, it is thought to arise from the odontogenic epithelium.^[5] It may occur intraosseously and extraosseously. The intraosseous variety is of two types, most common follicular type associated with an unerupted tooth and a less common extrafollicular type which is not related to an impacted tooth. All of our cases are of intraosseous type with one follicular variant and two extra follicular variants.^[8] In this case, the lesion appeared as a well-defined, unilocular radiolucency bound with an impacted tooth. There are some reported cases that did not show any resorption of the roots of teeth but in this case there was resorption of the roots of the deciduous lateral incisor.

AOTs are well encapsulated and have a benign biologic behavior. They never recur. The concept “envelopmental” given by Philipsen et al. cannot be ruled out.^[9] There have been a few cases of AOT-associated dentigerous cysts. An AOT readily separates from its bony crypt because it has a thick capsule and presents as an exophytic epithelial lining, which grows into the lumen. It can't be shunned when there is an impacted tooth, a dentigerous cyst develops first, followed by a cystic lesion, or it may directly develop as a cystic lesion.^[10] The AOT won't arise from the follicle of the tooth; rather, it arises from Hertwig's epithelial root sheath (HERS), which explains the tooth being completely within the lumen rather than the tooth root being within the bony crypt. AOTs are usually misdiagnosed as other odontogenic cysts and tumors on routine radiographic examinations. In this case, the orthopantomogram clearly shows the radiolucency covering the crown and extending apical to the cement-enamel junction, involving the entire root. Following surgical enucleation, the patient is healthy, and under follow-up

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