Clinicopathologic Case Report

Two sides of a coin: A report of two contrasting cases of adenomatoid odontogenic tumor with unusual presentations

ABSTRACT

This study aims to report two distinct cases of adenomatoid odontogenic tumor (AOT) with contrasting presentations. The first case involved a 12-year-old female patient with the presence of AOT in the posterior mandible extending into the mandibular ramus up till the coronoid process which is a rare reported site for AOT and the second case is of a 19-year-old male patient with the tumor in the anterior maxilla with an impacted central incisor. Both cases were successfully managed via surgical enucleation and are under regular follow-ups without any complication. The contrasting nature of the two clinical presentations with the same pathologic entity is reported here. Though AOT has been rarely reported extending into the mandibular ramus, a maxillofacial surgeon should be more circumspect of this pathology while dealing with benign swellings of the mandibular posterior region.

Keywords: Adenoameloblastoma, adenomatoid odontogenic tumor, mandible, maxilla, odontogenic tumor

INTRODUCTION

Adenomatoid odontogenic tumor (AOT) has a lengthy and voluminous history and available data which rarely fails to fascinate the radiologists, surgeons, and pathologists.[1] Steensland was the first to document AOT as "epithelioma adamantinum" in 1905 while Dreibladt in 1907 described it as "pseudo-adenoameloblastoma."[1-3] The name "adenomatoid odontogenic tumor" was proposed by Philipsen and Birn in 1969 which was adopted by the World Health Organization in 1971.[4,5]

AOT, the fourth most frequently occurring odontogenic tumor accounting for up to 3–7% of all odontogenic tumors,[3,6,7] is a benign, usually nonaggressive and noninvasive odontogenic epithelial tumor and is referred to as "two-third tumor" as in two-thirds of the cases, it occurs in young females, two-thirds of the cases arise in the maxilla, and two-thirds of the cases are associated with an unerupted canine. Since it occurs in the alveolar process of the jaws usually associated with an unerupted tooth having components of the dental lamina, enamel organ, reduced enamel epithelium, and/or their remnants, AOT is believed to originate from the odontogenic epithelium[7] although its origin is controversial.

This article reports two distinct cases of AOT with contrasting clinical presentations. One of them involved in an extremely unusual site, in the posterior mandible extending up till the coronoid process, while the other case involved anterior half of the palate associated with an impacted central incisor.

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Imran Khan, Anshul Singhal[1], Uqba Raza[2], Shubhangi Premchandani[2]

Department of Oral and Maxillofaial Surgery, [1]Consultant Maxillofacial Surgeon, Noida Dental Centre, Noida, Uttar Pradesh, [2]Faculty of Dentistry, Jamia Millia Islamia, New Delhi, India

Address for correspondence: Prof. Imran Khan, Room No. 509, Department of OMFS, Faculty of Dentistry, Jamia Millia Islamia, New Delhi - 110 025, India.
E-mail: ikhan3@jmi.ac.in

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CASE REPORTS

Case 1

A 12-year-old female patient accompanied by her parents presented with a complaint of intermittent discomfort and swelling on the left side of her lower jaw from last 3 months. The swelling had rapidly grown over the last 3 weeks to attain its present size. Medical history was non-remarkable. Extra-oral examination revealed a diffuse swelling measuring approximately 5 × 6 cm in size on the lower left side of face. On palpation, the swelling was mildly tender, incompressible, nonpulsating, and hard in consistency and was fixed to the underlying bone without any neurosensory deficit. Intraoral examination revealed a large swelling in relation to the left retromolar region. The left second molar was missing. On intraoral palpation, the swelling was found to be spherical, firm, immobile, and nontender, involving the body of the mandible extending up to the ascending ramus with a marked buccolingual cortical expansion. No discharge, bleeding, or foul smell was noted. The overlying skin was pinchable, nonerythematous, and locally afebrile.

An orthopantomograph (OPG) exhibited a well-demarcated osteolytic spherical radiolucent lesion with smooth and circumscribed margins [Figure 1a]; the pathology started from the left mandibular second molar region and extended posteriorly onto the mandibular ramus up till the coronoid process. The lesion appeared to displace the inferior alveolar canal (IAC), though the patient did not show any significant neurosensory alteration of lower lip or related structures. The eruption of mandibular second molar was hampered by the possible compression from the pathological lesion but the tooth itself was not contained within the lesion. A computed tomographic scan (CT scan) reconfirmed the extent of pathology with significant buccal cortical expansion and lingual plate perforation [Figure 1b and c]. Aspiration from the swelling did not yield any fluid. A differential diagnosis of a benign pathology such as ameloblastoma, odontogenic myxoma, and AOT was made after clinical and radiological assessment.

Under general anesthesia, the tumor was approached via intraoral vestibular incision and was enucleated and delivered in toto along with the second molar uneventfully [Figure 2a and b]. Intraoperatively IAC was identified and the nerve was preserved. Histopathologic analysis revealed an encapsulated, nodular tumor, with few slit-like cystic spaces at the periphery. The tumor cells were round to ovoid to spindle-shaped exhibiting cribriform pattern, gland formation and latticework fashion, some spindly area showed fasicular pattern with poor whorl formation, indicative of AOT [Figure 2c]. Post-operatively, the patient recovered well and has been under observation and regular follow-ups for last 1 year without any growth disturbances and complications [Figure 2d].

Case 2

A 19-year-old male patient reported with complaints of a slow-growing painless swelling in the right side anterior

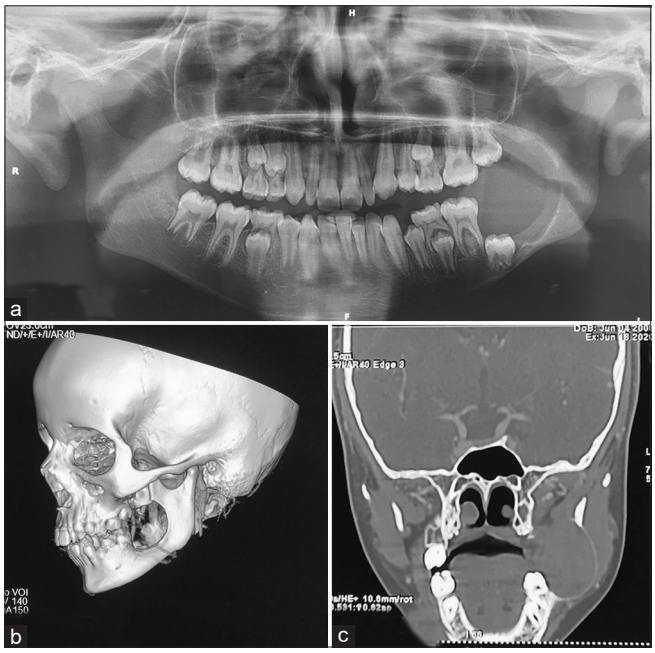

Figure 1: Orthopantomograph (OPG) and Computerised Tomographic Images (CT Images) showing the extent of the osteolytic lesion along with significant expansion and cortical perforation. (Case-1) (a-c)

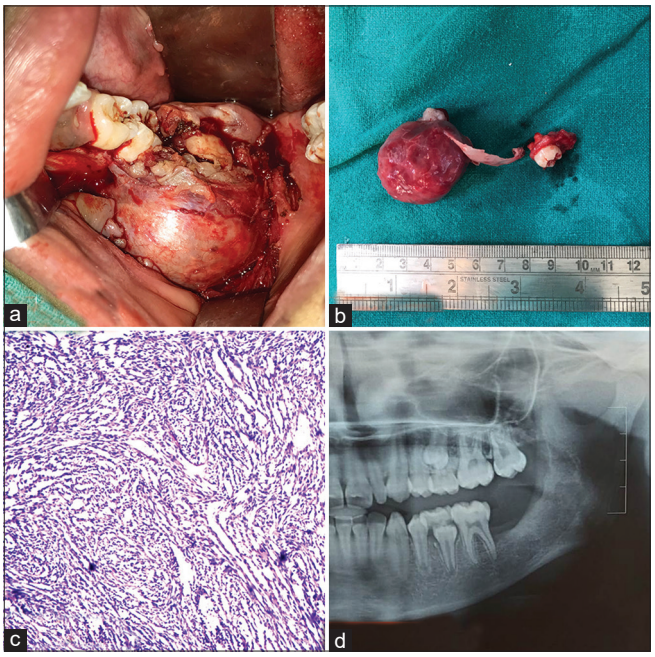

Figure 2: Intra operative picture depicting the size of the lesion as well as successful tumor enucleation. Histomicropraphic picture and six months post operative panoramic view. (Case-1) (a-d)

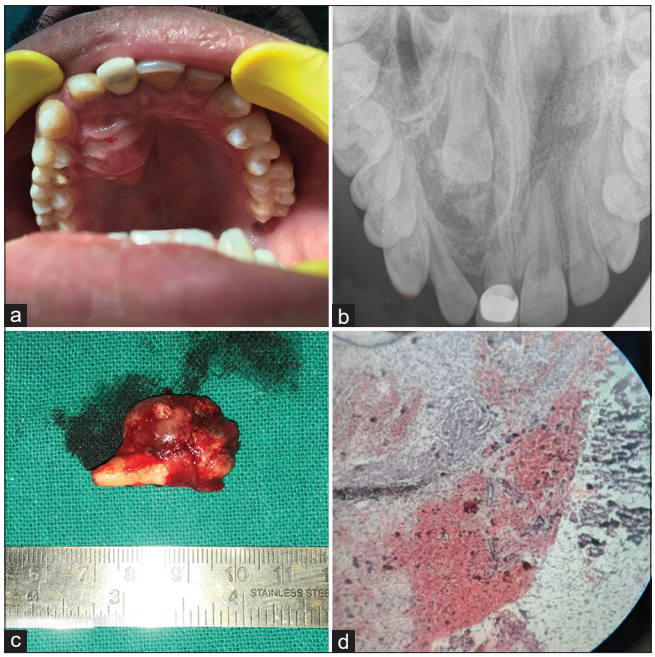

Figure 3: Clinical and Radiographic Picture depicting the pathology, Tumor enucleated in toto along with impacted maxillary central incisor, Histomicrographic image indicating towards AOT. (Case-2) (a-d)

palatal region from the last 5 years. There was no relevant medical history. Intraoral examination revealed swelling on the right side of anterior palate measuring approximately 4.5 × 2.5 cm and felt hard, firm, and immobile on palpation. Another relevant finding was a presence of an artificial crown on a possible deciduous central incisor.

Aspiration did not yield any fluid. A maxillary true occlusal radiograph revealed a spherical radiolucent lesion having specs of radio-opaque calcifications within the borders of the lesion was well circumscribed and radiopaque starting from the midline and extending till the second premolar/first molar region [Figure 3a and b]. As suspected, the permanent right central incisor was impacted and was visualized within the center of the lesion on the radiograph. A differential diagnosis of AOT, calcifying epithelial odontogenic tumor, was made based on radiographic findings. Under local anesthesia, the lesion was approached palatally and enucleated in toto along with the impacted central incisor without any intraoperative complications [Figure 3c]. Histopathological analysis was indicative of AOT [Figure 3d]. Postoperative recovery of patient was uneventful and the patient is under regular follow-up from 2 years without any negative outcomes.

DISCUSSION

Philipsen and Birn described the following variants of AOT as per its clinical and radiographic presentation:[2,4,6]

(1) Central (intraosseous)
   (a) Follicular type (70%)—the tumor is associated with an impacted tooth.
   (b) Extrafollicular type (25%)—not associated with an impacted tooth.
(2) Peripheral/Extraosseous (5%)—seen in the gingiva as a fibrous epulis

Histopathologically, AOT is usually encapsulated, composed of ovoid-to-polygonal-to-spindle-shaped cells forming sheets and whorls in a connective tissue stroma,[8,9] as evident in both our cases. In our first case (extrafollicular variety), the tumor was seen to comprehend the posterior mandible, extending into the ramus and involving the coronoid process; this location for AOT is very rare and has been sparsely reported in literature. The second case (follicular variety) had a typical clinical presentation of AOT (slow growing, painless, anterior palate involvement); still, involvement of impacted central incisor tooth within the lesion instead of a canine is a distinct feature. The present case report highlights contrasting nature of a pathologic entity with separate clinical presentations. As previously stated AOT in the mandibular posterior region has seldom been reported, while searching the published literature on various indexing database sites, only a few were found with extrafollicular variants of AOT in the posterior mandible.[10-12] Details of the same have been enlisted [Table 1]. Out of these three reports listed [Table 1], only one[12] reports the lesion to be extending up till the mandibular ramus involving the coronoid process.

Since the tumor is encapsulated, noninvasive and easily separable from the bone, enucleation and curettage are generally sufficient.[10-12] In both our cases, we could easily separate the tumor with the adjacent bone and enucleate it without any difficulties. Prognosis mostly is excellent, both of our cases have been on regular follow-up ever since without any complications.

CONCLUSION

AOT is an uncommon, benign, painless, odontogenic tumor of epithelial origin, very frequently misdiagnosed as odontogenic cyst. Interestingly, our present cases differ in their clinical presentations, yet the diagnosis remains the same. AOT is prevalent in maxillary anterior region as seen in our second case but its presence in the mandibular posterior region is quite unusual as in the first case. Furthermore, it might be fast growing as seen in our first case without any impacted tooth in the lesion (extrafollicular-type AOT) which is less common as compared to the follicular variety. Although AOT involving the mandibular posterior region is

Table 1: Published data reporting extrafollicular AOT extending in the posterior mandible

Author (s)	Country and year of publication	No. of cases	Patient age/sex	Location	Type of adenomatoid odontogenic tumor
Shivali *et al.*[10]	India 2013	1	18 Y/Male	Left body of the mandible region	Extrafollicular
Lang *et al.*[11]	Taiwan 2013	1	30 Y/Female (recurrent tumor, with ghost cells presence operated thrice, ultimately resection done)	Mandibular left posterior region	Extrafollicular
Ramlal *et al.*[12]	India 2010	1	18 Y/Female	Left mandibular ramus region	Extrafollicular

rare, careful case history, adequate interpretation of clinical, radiographic, and histological findings aid in arriving to the correct diagnosis.

Declaration of patient consent

The authors certify that they have obtained appropriate patient consent forms. In the form, the patient (s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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