

Up-to Date Review And Case Report

A case of large adenomatoid odontogenic tumor in the posterior region of the mandible showing root resorption

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Abstract – Introduction: Adenomatoid odontogenic tumor (AOT) is a rare tumor of epithelial origin, and usually presents as a unilocular radiolucency in the maxillary anterior region in adolescent females. **Observation:** A 31-year-old Japanese male, having a large adenomatoid odontogenic tumor from the right molar region to the left anterior region of the mandible showing root resorption of the neighboring teeth, was presented to the hospital. The lesion was totally resected under general anesthesia. **Commentary:** AOT may cause displacement of the neighboring teeth. But root resorption is a very rare finding. AOTs are relatively small in size. **Conclusion:** The patient was under follow-up and had not shown any signs of recurrence 12 months after surgery.

Introduction

Adenomatoid odontogenic tumor (AOT) is a rare tumor of epithelial origin accounting for between 2.2 and 7.1% of all odontogenic tumors [1]. AOT appears as an intraoral – extraoral swelling in the maxilla, and is sometimes referred to as “Two-thirds tumor” because it occurs in the maxilla in about 2/3 cases, about 2/3 cases in young females, 2/3 cases are associated with an unerupted tooth, and 2/3 affected teeth are canines [2]. AOT frequently resembles lesions like dentigerous cyst or ameloblastoma on radiographic finding. Intraosseous AOT may cause displacement of the neighboring teeth. Root resorption, which is frequently presented in ameloblastoma, is a very rare finding [3]. In cases reported till now, AOTs are relatively small in size and they do not exceed 1–3 cm in diameter [4]. We report a rare case of large adenomatoid odontogenic tumor from the right molar region to the left anterior region of the mandible showing root resorption.

Observation

A 31-year-old Japanese male was referred for further evaluation of a cystic radiolucent lesion of the mandible. He had no significant medical history and was not using any

medications. There was no anesthesia or paresthesia of the lower lip, chin, or jaw, and there was no history of trauma. On extraoral examination, a slight swelling on the right side of the mandible was recognized in comparison with the opposite side (Fig. 1). On palpation, there was neither local rise in temperature nor tenderness in the swelling region. Intraoral examination revealed painless bony swelling extending bilaterally to the buccal and lingual vestibule from the right mandibular first molar to the left mandibular lateral incisor. The teeth associated with the swelling – the incisors, canines, premolars and first molars of the right and right side of the mandible – gave a positive response to vitality tests. The right second premolar was detected tooth mobility. Panoramic radiograph showed a well-defined, unilocular radiolucent lesion in the mandible extending from the right first molar to the left lateral incisor. The large lesion was associated with the impacted and displaced right first premolar. There was deviation of the root of right second premolar and the root of right canine. Root resorption was also evident in relation to the right second premolar and the right first molar in both panoramic radiograph and periapical radiograph (Fig. 2A, 2B). The patient was further subjected to a computed tomography (CT) examination, which demonstrated a large expansive lesion contained multiple and minimal flecks of high density. The large lesion was associated with the impacted and displaced right first premolar. There was deviation of the root of right second premolar and the root of right canine similar to the

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Fig. 1. A slight swelling on the right side of the mandible in comparison with the opposite side.

panoramic findings. The dimensions of the lesion measured 37.0 mm × 27.0 mm × 17.8 mm (Fig. 3). Biopsy was performed under local anesthesia. Histopathologically, epithelial cells had outgrown as sheets, and irregularly calcified materials were present. Thus, the lesion was suspected as calcifying epithelial odontogenic tumor.

The lesion was totally resected with extraction of the impacted right first premolar and the right second premolar under general anesthesia. The mandible was exposed after a Wasmund-type incision and a medial and distal vertical incision in the buccal vestibule from the right first molar to the left lateral incisor. The buccal cortex was intact. After extraction of the right second premolar and osteotomy, a reddish, bulky layer of granular-like tissue became evident inside the bone cavity. The cavity filled solid tumor including the tooth. The tumor was consequently enucleated from the cavity. The tooth was attached loosely to the tumor and was removed easily (Fig. 4A). There were no apparent infiltrations of the surrounding bones and no exposure of the neighboring root surface in the bone cavity. Macroscopically, the mass was well encapsulated with cystic areas along with an embedded mandibular first premolar in the tumor mass (Fig. 4B). Histopathological examination revealed sheets, ducts (Fig. 5A: arrow), and whorls (Fig. 5B: arrow) of darkly staining ovoid to round epithelial cells suggestive of odontogenic epithelial cells. A few basophilic calcifications were also observed (Fig. 5C: arrow). The duct-like structures were lined by columnar cells (Fig. 5D: arrow).



Fig. 2. Panoramic radiograph showed a well-defined, unilocular radiolucent lesion in the mandible. The large lesion was associated with the impacted and displaced right first premolar (A). Root resorption was also evident in relation to the right second premolar and the right first molar (A and B).

Based on these findings, the lesion was histopathologically diagnosed as follicular type of AOT.

Healing of the wound was uneventful postoperatively. The patient was under follow-up and had not shown any signs of recurrence 12 months after surgery.

Commentary

Adenomatoid odontogenic tumor (AOT) is a rare tumor of epithelial origin accounting for between 2.2 and 7.1% of all odontogenic tumors and is given a ranking of fourth or fifth among the odontogenic tumors only surpassed by odontomas, myxomas, ameloblastomas and cemento-osseous tumors or lesions [1,5]. More than 95% of AOTs are intraosseous, but extraosseous variants have been documented. About three quarters of cases occur in a pericoronal relationship, which has led some authorities to subclassify the tumors as follicular or extrafollicular [6]. In this case, the lesion was diagnosed as the intraosseous/follicular type of AOT.

AOT sometimes referred to as “Two-thirds tumor”, because it occurs in the maxilla in about 2/3 cases, about 2/3 cases in young females, and 2/3 affected teeth are canines [2]. The follicular and extrafollicular variants together

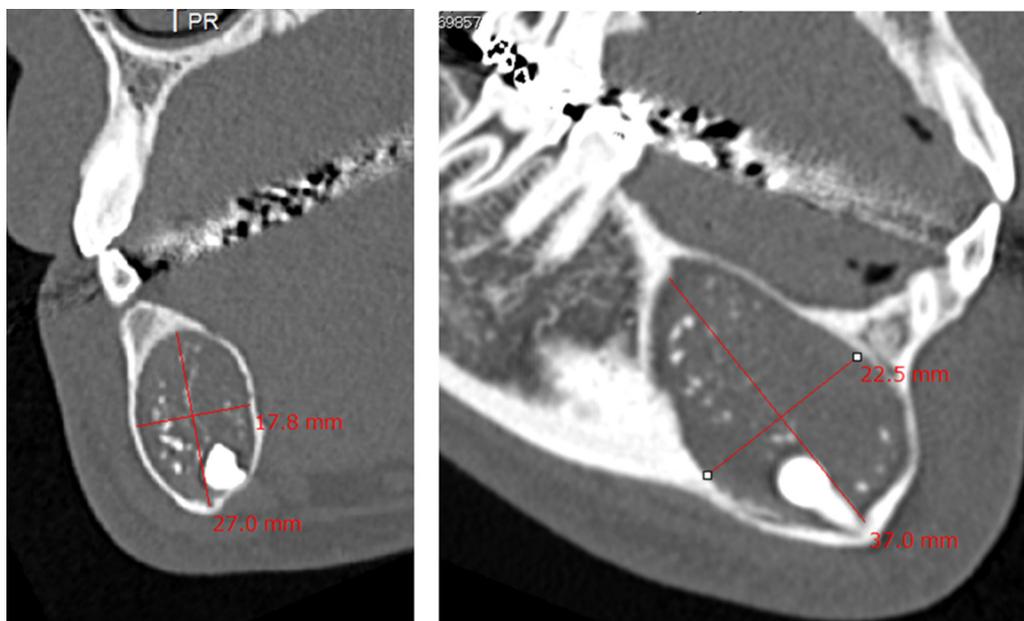


Fig. 3. Computed tomography (CT) imaging demonstrated a large expansive lesion contained multiple and minimal flecks of high density. The dimensions of the lesion measured 37.0 m × 27.0 mm × 17.8 mm.

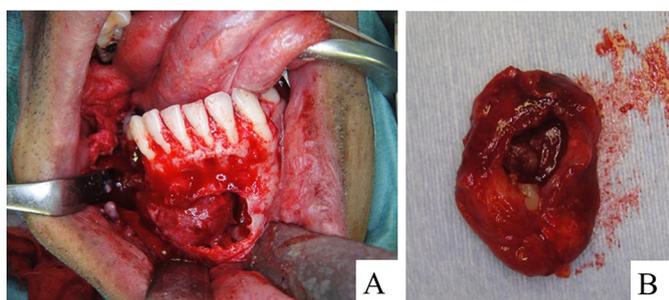


Fig. 4. (A) A reddish, bulky layer of granular-like tissue became evident inside the bone cavity. (B) The mass was well encapsulated with cystic areas along with an embedded mandibular first premolar in the tumor mass.

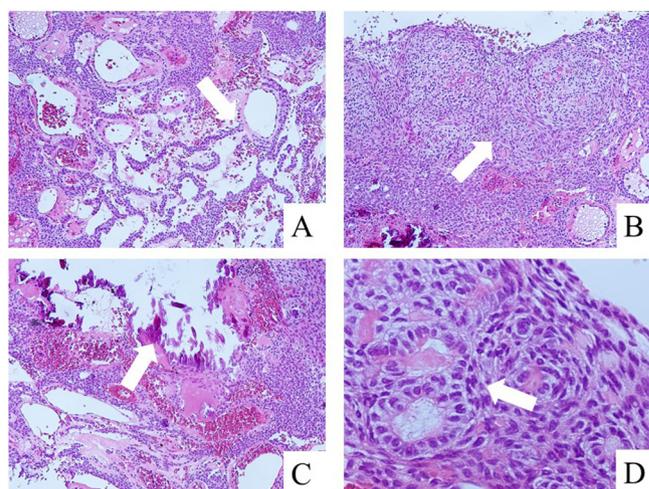


Fig. 5. Histopathological examination revealed sheets, ducts (A: arrow), and whorls (B: arrow) of darkly staining ovoid to round epithelial cells suggestive of odontogenic epithelial cells. A few basophilic calcifications were also observed (C: arrow). The duct-like structures were lined by columnar cells (D: arrow).

(approximately 96%) are more commonly found in the maxilla than in the mandible with a total ratio of 2.1:1 [1]. The female-male ratio for all age groups and AOT variants together-and globally-is 1.9:1 [1]. If cases reported from Sri Lanka and Japan are considered separately, they show a female: male ratio of 3.2:1 and 3.0:1, respectively [5,7]. The age range of patients with AOT varies between 3 and 82 years at the time of diagnosis [8]. No less than 68.6% of the tumors are diagnosed in the second decade of life and more than half of the cases (53.1%) occur within the teens (13 ± 19 years of age). This age distribution with a very tall peak in the second decade makes the AOT unique among odontogenic tumors [1]. Concerning the distribution of unerupted permanent teeth found in association with the follicular AOT, all four canines account for 59% and the maxillary canines alone for 40%. Unerupted first and second molars are the teeth most rarely involved in AOTs, only four cases having been reported so far. Association between central

AOT and unerupted deciduous teeth is exceedingly rare, only two cases having been published [6]. This report showed that AOT was present in the mandibular of 31-year-old man.

In our case, a radiographic diagnosis of AOT was arrived at considering the multiple scattered radiopaque flecks in the lesion associated with an unerupted impacted tooth. However, the large size of the lesion and root resorption of the neighboring teeth in our case is unusual in an AOT. Hence, a differential diagnosis of other lesions such as calcifying cystic odontogenic tumor was also considered. After biopsy, the

Table I. Seven cases of AOT with root resorption.

Author	Age	Sex	Site	Variant	Resorbed teeth
Narayanan <i>et al.</i> [3]	17 years	Female	Mandibula	Follicular	Premolars, Molars
Bartake <i>et al.</i> [11]	14 years	Female	Anterior maxilla	Follicular	Central incisor, Premolars
Dayi <i>et al.</i> [12]	15 years	Female	Anterior maxilla	Follicular	First premolar
Nomura <i>et al.</i> [13]	64 years	Male	Mandibula	Extrafollicular	Canine
Nigam <i>et al.</i> [14]	15 years	Female	Anterior maxilla	Follicular	Lateral incisor, First premolar
Chuan-Xiang and Yan [15]	16 years	Male	Maxilla	Follicular	Premolars
Saluja <i>et al.</i> [16]	17 years	Male	Mandibula	Follicular	Premolars, Molars
Fujita <i>et al.</i>	31 years	Male	Mandibula	Follicular	Premolars, Molars

lesion was suspected as calcifying epithelial odontogenic tumor. The lesion was finally diagnosed as follicular type of AOT. AOT frequently resembles lesions like dentigerous cyst or ameloblastoma. In fact, 77% of follicular type AOT is initially diagnosed as dentigerous cysts [9]. Gadewar and Srikant [10] reported a case of AOT having a large cystic space lined by thick stratified squamous epithelium and suggested that the presence of AOT in the nodules of cystic lining gives a histological proof that AOT had transformed from a cyst. In our case, the mass of the lesion was well encapsulated with cystic areas in the absence of stratified squamous epithelium, but the lesion mimicked an odontogenic cyst in radiographic findings.

Intraosseous AOT may cause displacement of the neighboring teeth. Root resorption, which is frequently presented in ameloblastoma, is a very rare finding [3,6]. To the best of our knowledge, 7 cases of AOT with root resorption have been reported as Table I [3,11–16]. Struther and Shear [17] reported that the incidence of root resorption in association with ameloblastomas (81%) is far higher than that associated with the cystic lesions (0–55%). Calcifying epithelial odontogenic tumor, as which our case was suspected after biopsy, is less aggressive than ameloblastoma. Root resorption is reported as a rare finding in calcifying epithelial odontogenic tumor (4%), unlike solid ameloblastoma [18].

AOTs are relatively small in size. Usually, they do not exceed 1–3 cm in diameter [4]. The case of AOT we experienced was large size from the right molar region to the left anterior region of the mandible.

Although some AOT with odontoma are large lesions, their clinical behavior seems similar to conventional AOT. None of the cases showed recurrence, suggesting that the tumor can be treated conservatively. A recurrence case of AOT was reported by Chuan-Xiang and Yan [15]. We should be careful to follow up cases of AOTs after surgery.

Conclusions

We report a rare case of large AOT from the right molar region to the left anterior region of the mandible showing root

resorption. The patient was under follow-up and had not shown any signs of recurrence 12 months after surgery. Careful follow-up examinations should be conducted in this disease.

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