

Adenomatoid odontogenic tumors: an analysis of 67 cases in a Thai population

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Objective. The aim of this study was to determine the biologic profile of the adenomatoid odontogenic tumor (AOT) in a Thai population.

Study design. Sixty-seven cases of AOT registered from January 1974 to May 2006 were studied retrospectively. Age, sex, variants of AOT, site and extent of tumors, associated impacted teeth, initial clinical diagnoses, clinical presentations, and duration of symptoms were analyzed.

Results. All AOT cases were intraosseously located, of which 55.2% were follicular and 44.8% extrafollicular. The 2 variants together were found more frequently in the maxilla than in the mandible, with a ratio of 2:1. Females were affected more often than males, with a ratio of 1.8:1. The peak incidence (56.7%) was found in the second decade of life. The majority of AOT lesions (68.7%) was found in the anterior jaws. Adenomatoid odontogenic tumors were observed more often on the left side of jaws (50.7% vs. 38.8%). Canines were the most common teeth associated with follicular AOT (67.5%), and the maxillary canines alone accounted for 51.3% of all cases. The majority of our patients presented with a painless swelling and with duration of symptoms of 6 months and longer.

Conclusions. The distribution and characteristics of AOT in a Thai population are similar to the findings found in other populations. Interestingly, in this series we observed a case involving an impacted deciduous canine and a case associated with an impacted maxillary third molar. (*Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2008;105:210-5)

The adenomatoid odontogenic tumor (AOT) was first reported by Harbitz¹ in 1915 under the name of *cystic adamantoma*. A multitude of authors during the early 1900s have given credit to researchers like Steensland,² Dreyblatt,³ and James and Forbes⁴ for reporting the first case of AOT. However, the first case that showed irrefutable proof of an AOT based on clinical features as well as microscopic findings was case III in Harbitz's report.¹ Philipsen and Birn⁵ proposed the widely accepted and currently used name *adenomatoid odontogenic tumor*, a term that was adopted by the first edition of the World Health Organization classification of odontogenic tumors in 1971. Adenomatoid odontogenic tumor is defined as a tumor composed of odontogenic epithelium in a variety of histoarchitectural patterns, embedded in a mature connective tissue

stroma, and characterized by slow but progressive growth.⁶

Adenomatoid odontogenic tumor has been described as an uncommon benign odontogenic tumor with a relative frequency (RF) of 2.2% to 7.1%.⁷⁻⁹ However, a very recent multicenter study¹⁰ covering 58 publications has disclosed that the RF in fact varies from 0.6% to 38.5% worldwide, with the 2-digit figures being reported from the African continent and from Nigeria in particular. Adenomatoid odontogenic tumor is mostly encountered in young patients, especially in the second decade of life, and is uncommon in patients older than 30 years of age. Females are afflicted by AOT more often than males.^{7-9,11-14} Maxilla is the predilection site of occurrence, almost twice as often as the mandible, and the anterior part of the jaw is more frequently involved than the posterior part. An unerupted maxillary canine is the tooth most commonly associated with AOT.^{7,14} Clinically, AOT presents as a slow-growing symptom-free lesion and is frequently discovered during routine radiographic examination.¹⁵

Adenomatoid odontogenic tumor can occur both intraosseously and extraosseously. The intraosseous AOT may be radiographically divided into 2 types: follicular (or pericoronal) and extrafollicular (or extracoronal) types. The former is characterized by a well-defined

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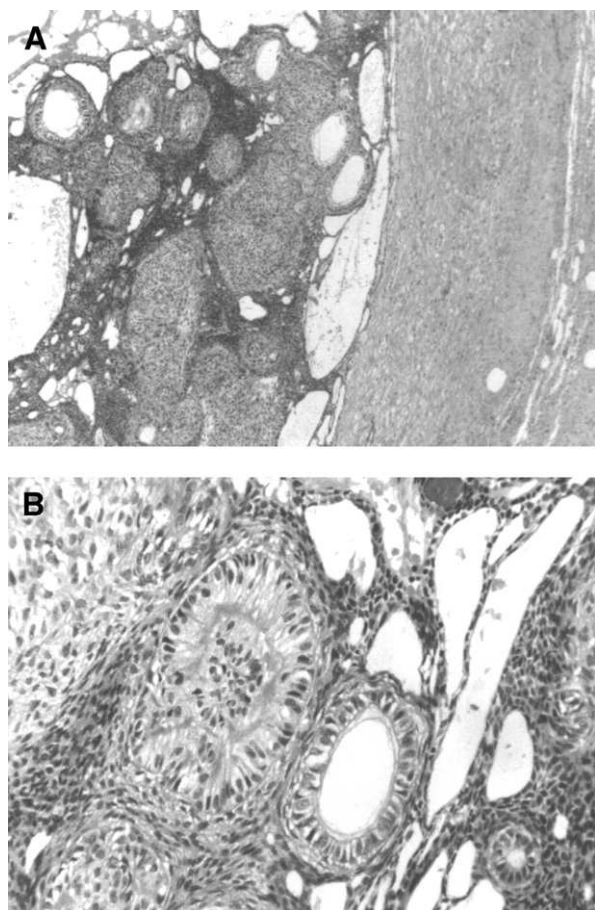


Fig. 1. Photomicrographs of adenomatoid odontogenic tumor in an 18-year-old female. **A**, Low-power view showing a well-developed connective tissue capsule surrounding the tumor (hematoxylin-eosin, original magnification $\times 20$). **B**, High-power view showing the characteristic ductlike structures, lined by a single row of columnar epithelial cells. (hematoxylin-eosin, original magnification $\times 50$).

unilocular radiolucency surrounding the crown and is often part of the root of an unerupted tooth. The latter is likewise a well-defined radiolucency but located between, above, or superimposed upon the root of an unerupted tooth.⁷ Minute, variable-shaped radiopacities are frequently found within the lesion. The extraosseous, peripheral, or gingival types of AOT are rarely detected radiographically, but there may be slight erosion of the underlying alveolar bone cortex.^{7,14}

Adenomatoid odontogenic tumor is usually surrounded by a well-developed connective tissue capsule (Fig. 1, A). It may present as a solid mass, one large cystic space, or as numerous small cystic spaces. The tumor is composed of spindle-shaped cells or polygonal cells forming sheets and whorled masses in a scant connective tissue stroma. Between the epithelial cells

as well as in the center of the rosettelike structures are amorphous eosinophilic materials. Anastomosing cords or strands of epithelium that are 1- or 2-cells thick are seen at the periphery of some tumors. The characteristic ductlike structures are lined by a single row of columnar epithelial cells, the nuclei of which are polarized away from the central lumen. The lumen may be empty or contain amorphous eosinophilic material (Fig. 1, B). Dystrophic calcification in varying amounts and in different forms is usually encountered in most AOTs within the lumina of the ductlike structures, scattered among epithelial masses or in the stroma.^{7,9,15} Dentinoid material or osteodentin has been reported by several authors, whereas occurrence of enamel matrix is extremely rare. Some cases of AOT demonstrate areas resembling other odontogenic tumors such as calcifying epithelial odontogenic tumor, odontoma (and similar tumors), and calcifying odontogenic cyst (calcifying cystic odontogenic tumor). They should be classified as histologic variants of AOT,^{7,12,15} as they have been shown not to influence the biologic behavior of AOT.

There were few reports on AOT in Asian populations, thus our analysis of AOT in a Thai population could provide useful additional data to the literature. The aim of this study was to determine the biologic profile of AOT in a Thai population.

MATERIAL AND METHODS

The histopathologic records of the Department of Oral Pathology, Chulalongkorn University, were reviewed for AOT lesions registered from January 1974 to May 2006. One investigator reevaluated the hematoxylin and eosin stained sections, and the histopathologic diagnosis was made according to the WHO Classification of Tumours.⁶ Data collected included age, sex, variants of AOT, site and extent of tumors, associated impacted teeth, initial clinical diagnoses, clinical presentations, and duration of symptoms. With regard to the site of occurrence, the jaw was divided into 2 areas: anterior region (from canine to canine) and posterior region (posterior to the canine).

Statistical analysis

Data was analyzed using SPSS for Windows (version 11.0; SPSS Inc., Chicago, IL). The chi-square test and *t* test were used to test the significance between groups. The level of significance was set at $P < .05$.

RESULTS

The patients were between 6 and 51 years of age at time of diagnosis, with a mean age of 21.1 ± 10.0 years. The mean ages of females and males were not different (20.9 ± 8.3 years and 21.4 ± 12.7 years,

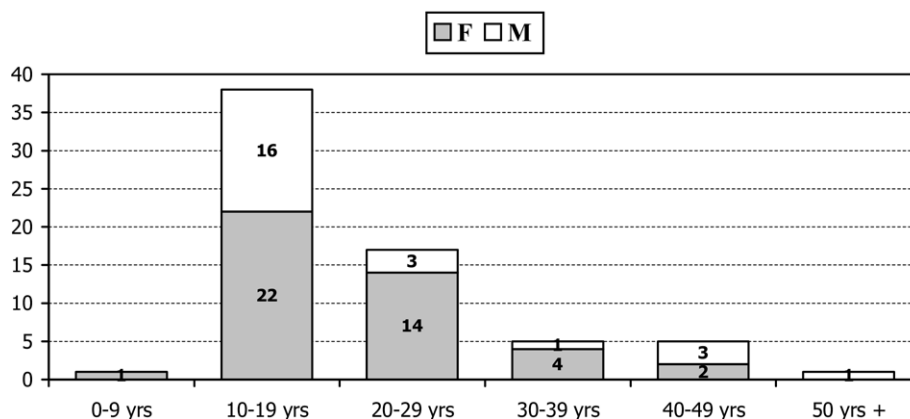


Fig. 2. Distribution of patients with adenomatoid odontogenic tumor (AOT) according to age and sex.

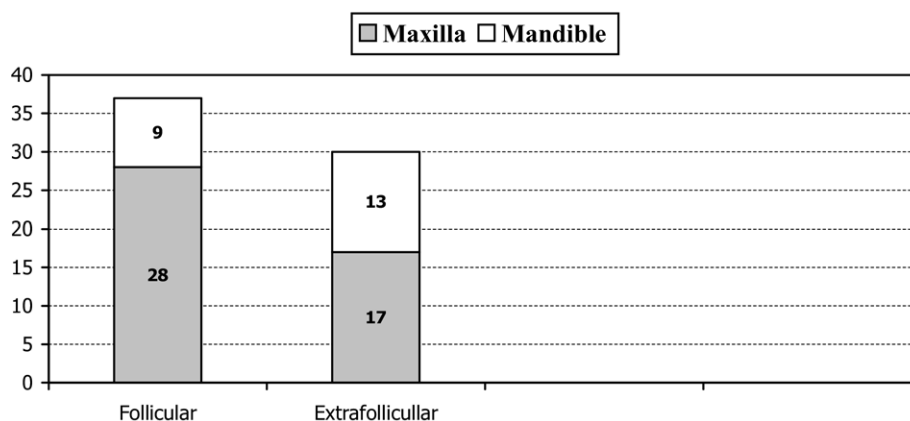


Fig. 3. Distribution of AOT variants according to location.

respectively; $P = .738$). The peak incidence of AOT (56.7%) was found in the range of 10 to 19 years of age (Fig. 2).

Of the 1268 cases of odontogenic tumors diagnosed, 67 AOTs were identified (RF = 5.3%). All cases were intraosseous, of which 37 (55.2%) were follicular and 30 (44.8%) were extrafollicular. The 2 variants together were found more frequently in the maxilla than in the mandible, with a ratio of 2:1 (Fig. 3). Females ($n = 43$) were affected more than males ($n = 24$), with a ratio of 1.8:1. In follicular AOT, the female to male ratio was 1.6:1, whereas it was 2:1 in the extrafollicular variant.

Table I shows the location of the 67 AOTs. Adenomatoid odontogenic tumor lesions were found more frequently in the anterior jaws (68.7%). There were more AOTs found on the left side than the right side of the jaw (50.7% vs. 38.8%). The remaining (10.5%) cases extended across the midline of the maxilla or the mandible.

Table II shows age of patients according to AOT variants and sex. Patients with the follicular type were

Table I. Location of 67 AOTs

Location	A n (%)	P n (%)	Total n (%)
Maxilla	32 (47.8)	13 (19.4)	45 (67.2)
Mandible	14 (20.9)	8 (11.9)	22 (32.8)
Total	46 (68.7)	21 (31.3)	67 (100.0)

AOT, adenomatoid odontogenic tumor; A, anterior region (from canine to canine); P, posterior region (posterior to the canine).

significantly younger at the time of diagnosis than those with the extrafollicular type (17.1 ± 6.9 years vs. 26.0 ± 11.0 years, respectively; $P < .001$). With regard to variants of AOT, there was no age difference between males and females.

Table III shows the frequency distribution of impacted teeth in the follicular AOT according to tooth groups and jaws. Of 37 cases of follicular AOT, 35 were associated with 1 tooth and the remaining 2 cases

Table II. Age of patients according to AOT variants and sex

AOT variants	Age range (y)	Age (y) Mean \pm SD		
		Females	Males	Total
Follicular (n = 37)	6-43	18.4 \pm 8.2*	14.7 \pm 3.0*	17.1 \pm 6.9 [‡]
Extrafollicular (n = 30)	12-51	23.8 \pm 7.5 [†]	30.7 \pm 15.3 [†]	26.0 \pm 11.0 [‡]
Total (n = 67)	6-51	20.9 \pm 8.3	21.4 \pm 12.7	21.1 \pm 10.0

AOT, adenomatoid odontogenic tumor.

*P = .105.

†P = .119.

‡P < .001.

Table III. AOTs associated with impacted teeth

Impacted teeth	Maxilla n (%)	Mandible n (%)	Total n (%)
Canines	17 (46.0)*	6 (16.2)	23 (62.2)
Lateral incisors	5 (13.5)	0 (0.0)	5 (13.5)
Central incisors	1 (2.7)	1 (2.7)	2 (5.4)
First premolars	2 (5.4)	2 (5.4)	4 (10.8)
Canine/first premolar	1 (2.7)	0 (0.0)	1 (2.7)
Lateral incisor/canine	1 (2.7)	0 (0.0)	1 (2.7)
Molar	1 (2.7)	0 (0.0)	1 (2.7)
Total	28 (75.7)	9 (24.3)	37 (100)

AOT, adenomatoid odontogenic tumor.

*Included 1 case of deciduous canine.

were associated with 2 teeth. Canines were the most common teeth associated with the follicular AOT (67.5%), and the maxillary canines accounted for 51.3% of all cases. The other teeth involved were incisors (21.6%), premolars (13.5%), and molar (2.7%). Moreover, there was 1 case with the maxillary left third molar and 1 case with the maxillary left deciduous canine involved in the AOT.

Table IV presents the initial clinical diagnosis of AOT cases. The diagnosis was specified in 64 cases. The most common diagnosis was dentigerous cyst, followed by AOT, calcifying cystic odontogenic tumor, and ameloblastomas.

Table V shows the clinical presentations in AOT patients. Most patients presented with painless swelling, whereas few patients exhibited pain or pain and swelling. Among patients with swelling, 5 patients had additional symptoms of nasal blockage, nasal bleeding, and exudate. There were 3 patients who had no symptoms, the lesions being detected by chance on radiographs.

The duration of symptoms was known in 49 cases and ranged from 1 week to 4 years. Of the 49 cases,

Table IV. Initial clinical diagnosis of AOTs

Initial clinical diagnosis	n (%)
Dentigerous cyst	18 (26.8)
Adenomatoid odontogenic tumor	11 (16.4)
Calcifying cystic odontogenic tumor	11 (16.4)
Ameloblastomas	9 (13.4)
Radicular cyst	3 (4.5)
Globulomaxillary cyst	2 (3.0)
Ossifying fibroma	2 (3.0)
Lateral periodontal cyst	1 (1.5)
Keratocystic odontogenic tumor	1 (1.5)
Calcifying epithelial odontogenic tumor	1 (1.5)
Miscellaneous	5 (7.5)
Nonspecified	3 (4.5)

AOT, adenomatoid odontogenic tumor.

Table V. Clinical presentations of AOTs

Presentations	n (%)
Painless swelling	56 (83.6)
Pain and/or swelling	7 (10.4)
Finding on radiographs	3 (4.5)
Not specified	1 (1.5)

AOT, adenomatoid odontogenic tumor.

28 patients reported duration of 6 months and longer (Fig. 4).

DISCUSSION

Adenomatoid odontogenic tumor is not a common odontogenic tumor, and it takes time to collect sufficient numbers of representative cases. Therefore, we have only 67 cases on record at the Department of Oral Pathology, Chulalongkorn University, for the past 31 years.

The results of our study agree to a large degree with most studies around the world.^{9,16,17} The RF of AOT among odontogenic tumors in the present study (5.3%) was in the range of previous reports. In comparison with Asian countries, our RF was lower than those reported for China (8.3%)¹⁸ and Sri Lanka (8.6%),¹⁹ but higher than those reported for Malaysia (0.3%)²⁰ and Hong Kong (3.6%)²¹ studies.

In our series, all tumors were intraosseously located, and the majority was follicular AOTs as previously described,^{14,16} with a strong female predilection.^{7-9,11-14} In support of previous reports,^{11,16,22} we found the highest incidence in the second decade of life. This study, in agreement with most reports, showed a predilection of AOT for the maxilla.^{11,13,14,16,17,19,21,22} However, some investigators have reported differently. Siar and Ng²⁰ and Fernandez et al.²³ showed a slight predilection for the mandible in their subjects. Arotiba et al.¹⁷ also

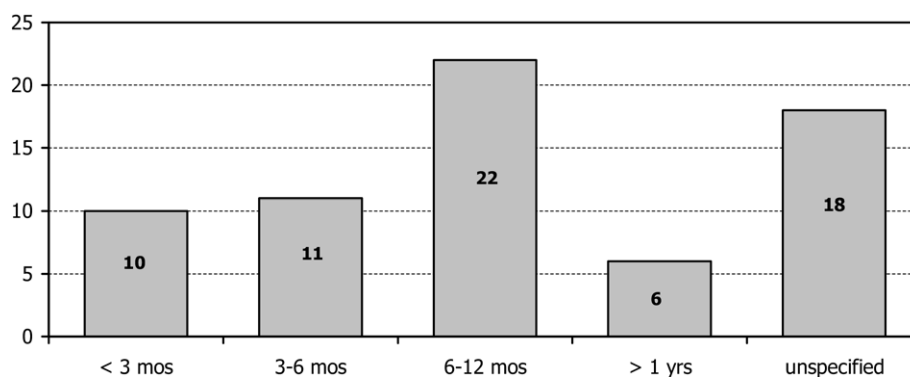


Fig. 4. Duration of symptoms in AOTs.

reported that extrafollicular tumors were more commonly found in the mandible in their study conducted in Africa.

Our patients with follicular AOT were also significantly younger than those with the extrafollicular variant.^{16,17} Our study detected no age-related differences between males, and females presented with a different type of AOT. This is unlike Arotiba et al.,¹⁷ who reported that males with extrafollicular tumors were relatively younger than their female counterparts.

The incidence of impacted teeth associated with AOT in this sample was comparable with most studies. Canines were the most common AOT-associated impacted teeth, of which the maxillary canines were the most common teeth involved.^{14,16,17,22} Interestingly, we had a case of a 43-year-old female who had the impacted maxillary left third molar associated with the AOT lesion. Unerupted molars were rarely involved in the AOT.^{16,24} Philipsen et al.¹⁰ suggested that tooth impaction and associated tumor development was likely to be diagnosed later in life when located in the posterior rather than the anterior jaw regions.

Most AOTs in our series were found in the anterior jaw regions; this finding was similar to other studies reporting anterior jaws as the preferable site for AOT lesions.^{14,17,19, 25,26} We observed a left side preference with a ratio of 1.3:1.

Dentigerous cyst was the most frequent initial clinical diagnosis of AOT in our study as well as in others.^{14,19,20,22} This is not surprising because the follicular AOT usually presents radiographically as a well-defined unilocular radiolucency surrounding the crown and part of the root of an unerupted tooth, resembling a dentigerous cyst. The initial clinical diagnosis as an AOT was correct in only 11 cases.

The most common clinical presentation of our patients was swelling, with a few having pain, which is similar to other previous reports.^{11,14,15,22} The majority

of our patients reported the duration of symptoms as 6 months and longer. This finding corresponds with the behavior of AOT—a slowly growing lesion that rarely causes pain—which is why patients rarely detect any changes at an early stage of tumor growth.

All cases in our series were treated by surgical enucleation, and no recurrences have been reported. This finding confirms the rarity of AOT recurrence.^{7,14,16,17,19,20}

In conclusion, we observed the similarities of the biologic profile of AOT between our study and previous studies. In this series, we observed a case involving an impacted deciduous canine and a case associated with an impacted maxillary third molar.

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