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Original Article

Treatment for large adenomatoid odontogenic tumors: clinical analysis of 13 cases

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Abstract: Adenomatoid odontogenic tumors (AOT) are exceptionally rare, accounting for 2.2-7.1% of all odontogenic tumors. The present study reviewed incidence, treatment, clinical features, and radiographic features of AOT patients and proposed an alternative treatment for large AOT. These AOT patients were admitted between June 2006 to June 2017 and retrospectively reviewed. Gender, age, lesion site, affected tooth, types, and treatment of these patients were recorded. A total of 13 AOT patients were documented. Age range was 11 to 65 years, with a mean age of 21.15 years. High incidence was found in the second decade of life (8 cases; 66.67%). Females were affected more than males, with a ratio of 1.6:1. The maxilla had 6 cases (46.15%) and the mandible had 7 cases (53.85%), with a ratio of 1:1.17. Moreover, 10 follicular cases (76.92%), 3 extra-follicular cases (23.08%), and 0 extra-osseous cases were found. In follicular AOT, the most commonly impacted tooth was canine (8; 80.00%). A total of 10 cases (83.33%) were treated with enucleation, 2 cases (16.67%) were treated with fenestration decompression, and 1 case was not treated due to poor general conditions. After two years of follow-ups, 1 (8.3%) case had a relapse. There were no clear signs of recurrence in the remaining cases. In conclusion, AOT was more common in females, most in the second decade of life, usually associated with canines. Conservative surgical enucleation is the treatment for AOT. However, for large AOT, fenestration decompression may be an effective treatment.

Keywords: Adenomatoid odontogenic tumors, fenestration decompression, enucleation

Introduction

Adenomatoid odontogenic tumors (AOT) are exceptionally rare and benign, originating in the odontogenic epithelium and accounting for 2.2-7.1% of all odontogenic tumors that include a mass [1-4]. It was first introduced by Dreilbaldt in 1907, as a pseudoadenoameloblastoma, and first reported by Harbitz in 1915, as a cystic adamantinom [1, 5, 6]. The term “adenomatoid odontogenic tumor” was proposed in 1969 by Philipsen and Birn. It was adopted by the World Health Organization for its classification of odontogenic tumors [5, 7-9]. Adenomatoid odontogenic tumors are mainly seen in the maxillary and more frequently in young female patients. AOTs usually present as asymptomatic swelling, slowly growing and often associated with an unerupted tooth. Unerupted permanent canines are the teeth most often involved in AOTs [1, 3, 9]. Lesions are often discovered incidentally upon X-ray examination. Radiographically, AOT is a well-defined unilocular lesion surrounding the crown of an unerupted tooth, often misdiagnosed as odontogenic cysts, such as dentigerous cysts and ameloblastomas [3, 10]. Diagnosis of AOT usually depends on pathological features. Due to tumor growth, the adjacent teeth can be displaced, but teeth root resorptions are rare. Recurrence of AOT is exceptionally rare, therefore the prognosis is better.

The most commonly applied treatment for AOT is conservative enucleation and curettage [11]. However, for large AOT, this treatment can lead to some complications, such as mandibular fractures, mandibular continuity defects, maxillary sinusitis, and damage of infraorbital or inferior alveolar nerves. Fenestration decompression procedures have achieved good results in various types of large odontogenic cysts, with high success rates [12, 13]. This treatment creates an opening to reduce pressure within a

Table 1. Details of included patients

No	Gender	Age (years)	Site	Involved tooth	Types	Treatment
1	Female	13	Maxilla	No	Extrafollicular	Enucleation
2	Male	15	Maxilla	23	Follicular	Enucleation
3	Female	20	Maxilla	13	Follicular	Enucleation
4	Female	22	Maxilla	22	Follicular	Enucleation
5	Male	65	Maxilla	13	Follicular	No surgery
6	Male	23	Mandible	No	Extrafollicular	Enucleation
7	Female	32	Mandible	33	Follicular	Enucleation
8	Female	13	Mandible	43	Follicular	Decompression
9	Female	15	Mandible	32	Follicular	Enucleation
10	Male	19	Mandible	43	Follicular	Enucleation
11	Male	13	Mandible	No	Extrafollicular	Enucleation
12	Female	11	Mandible	43	Follicular	Enucleation
13	Female	14	Maxilla	23	Follicular	Decompression

cystic cavity, inducing bone formation. For this reason, many surgeons prefer these methods for treating large cysts. It has been considered whether fenestration decompression can be used as treatment for large AOT. Therefore, the present study reviewed cases of adenomatoid odontogenic tumors over the last 10 years, reporting a successful case of AOT treated with fenestration decompression.

Materials and methods

This retrospective study was performed using the database of patients of the Department of Oral and Maxillofacial Surgery, Nanjing Stomatological Hospital, Medical School of Nanjing University. Between June 2006 to June 2017, a total of 13 patients were diagnosed with adenomatoid odontogenic tumors (AOT), according to the WHO Classification of Tumors. This study analyzed gender, age, site of the lesion, affected tooth, types, and treatment methods in these patients. Detailed data of the 13 patients are shown in **Table 1**.

Results

There was a total of 13 patients examined, with 8 females and 5 males, respectively, with patient ages ranging from 11 to 65 years. Mean age was 21.15 years. Females were affected more than males, with a ratio of 1.6:1. In addition to a 65-year-old patient, high incidence was found in the second decade of life (8 cases; 66.67%). Data that was found in the study was comparable to that stated in the literature of A. Mohamed [1, 4]. A total of 6 (46.15%) AOTs occurred in the maxilla and 7 (53.85%) in the mandible, with a ratio of 1:1.17. All AOTs presented in this study were intraosseous. Intraosseous AOT is radiographically divided into 2 types: follicular and extrafollicular. A total of 10 (76.92%) AOTs were follicular while 3 (23.08%) AOTs were of extrafollicular. There were no extraosseous/gingival AOT types in the cases collected for this article. In follicular AOT, the most commonly impacted tooth was the canine (8; 80.00%), followed by the lateral incisor (2; 20.00%). Radiographically, the imaging manifestation of follicular AOT is a cyst-like appearance, a distinct border of the unilocular radiolucency has been associated with unerupted tooth and scattered miliary calcifications. Extrafollicular AOT were not involved in the unerupted tooth.

Moreover, 10 cases (83.33%) were treated with enucleation, 2 cases (16.67%) were treated with fenestration decompression, and 1 case was not treated with surgery methods due to poor general conditions. After two years of follow-ups, 1 (8.3%) case had a relapse. There were no clear signs of recurrence in the remaining cases. After six months of decompression, the No 8, 13 patient underwent surgical enucleation of the lesion.

Herein, a case of large AOT in a 13-year-old young girl is presented.

Case report

A 13-year-old girl presented in October 2010 with a progressively enlarging bulge of the right mandible for 2 years. She had no other chief complaint symptoms, such as pain and numbness of the lower lip. She had no noteworthy family history of this condition. Clinical examination revealed a large right facial mass localized to the mandible (**Figure 1**). Intraoral examination revealed a smooth bulge in the right mandible alveolar process extending to the inferior margin of mandible. There was the sense of table tennis on palpation. The right mandibular permanent canine was absent,

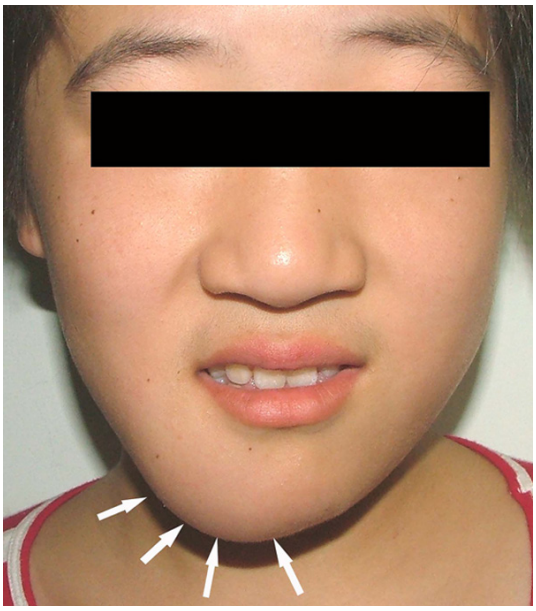

Figure 1. Before therapy, the patient showed facial asymmetry with the right mandible showing greater bulging.

Figure 2. Before therapy, panoramic radiograph showed a unicystic radiolucen lesion in the mandible with a comparatively clear demarcation. Tooth 43 was located on the floor of this lesion. There was no resorption of the root apices.

while the right mandibular premolar, lateral incisor, and incisor were displaced. There were no palpable cervical or submandibular lymph nodes and the chest radiograph was normal. Preoperative panoramic radiographs showed a unilocular well-circumscribed radiolucency extending from the first molar of the right mandible to the canine of the left mandible. It was approximately 5 cm in length and tooth 43 was located on the floor of this lesion. The neighboring teeth were displaced, but there was no resorption of the root apices (**Figure 2**). Computed tomography (CT) scans revealed an extensive expansile osteolysis with stippled calcifications (**Figure 3**). According to radiologic and clinical findings, diagnosis of AOT was made. Due to the massive extension of the lesion, an incisional biopsy was performed under local anesthesia. The lesion was decompressed between the mandibular first premolar and lateral incisor. Histopathological evaluation of the lesion confirmed the diagnosis of AOT (**Figure 4**). An individual obturator was made with acrylic resin (**Figure 5**). After treatment, patients were instructed how to wear and remove the obturator and they were informed to clean the obturator three times a day using saline solution to avoid obstruction and infections. The patient was scheduled for radiographic examination and pulp vitality test follow-ups after an interval of three months. Six months post decompression, the diminished lesion was enucleated completely under general anesthesia with an intraoral approach. After the treatment process, the pulp vitality of the teeth involved were still normal and the teeth involved were preserved with no need for root canal therapy. No bone graft was placed in the cavity. The impacted mandibular canine was extracted. The postoperative course was uneventful and no complications occurred (**Figures 6**, **7**). There were no signs of recurrence at the follow-up after 50 months.

Discussion

Adenomatoid odontogenic tumors (AOT) are benign slowly progressing growths that account for 2.2-7.1% of all odontogenic tumors. These originate in the odontogenic epithelium, such as dental lamina or its remnant [1-3, 14, 15]. Incidence of AOT varies among countries in the world. This may be related to factors such as regional, ethnic, and medical levels. However, overall, AOT has low incidence. AOT often presents as a painless swelling of the jaw, usually occurring in the anterior region of the maxilla, approximately twice as often in females as in males. Most patients are in the second decade of life [16]. The patient in the present report was in the second decade and females were affected more than males, with a ratio of 1.6:1, in accord with previous literature. In this study, 6 (46.15%) AOTs occurred in the maxilla and 7 (53.85%) in the mandible, with a ratio of 1:1.17. The ratio was different from what was reported by Philipsen [4], due to an insufficient number of cases. AOT is commonly associated with

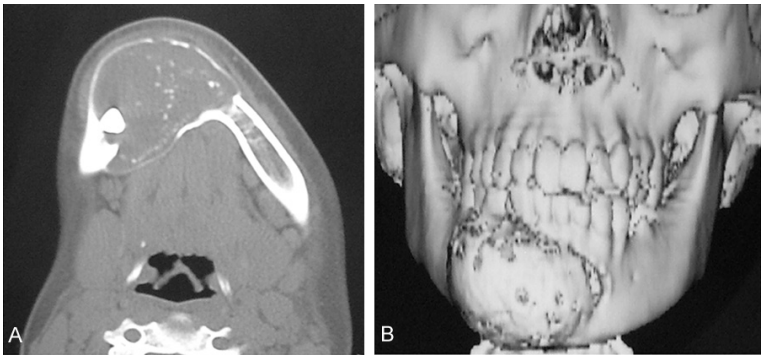

Figure 3. Computed tomography (CT) scans revealed an extensive expansile osteolysis with stippled calcifications.

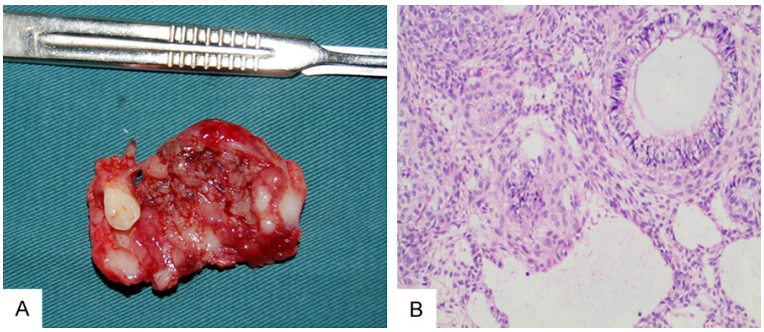

Figure 4. Lesions were surgically resected (A) and postoperative pathological findings displayed glandular duct-like structures consisting of cuboidal or low columnar cells (B).

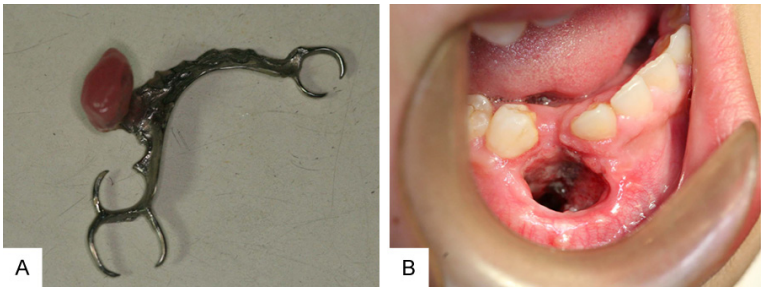

Figure 5. An individual obturator (A) was made with acrylic resin according to the opening size of the wound (B) and the obturator need regular adjustment.

unerupted teeth, especially maxillary canines [3, 10, 17]. The case reported in this study was associated with a mandibular unerupted canine. Similar AOT cases are relatively unique and rare. These tumors are slow growing and asymptomatic. They are often discovered incidentally with X-ray examinations. Due to the slow growth and generally asymptomatic characteristics of AOT, most patients are subjected to a bulge for years until it produces significant or obvious deformity and discomfort.

There are two major types of AOT, intraosseous and extraosseous. Intraosseous AOT is divided into 2 variants: follicular and extrafollicular [1, 4, 9, 10]. Radiographically, the manifestation of intraosseous AOT is a cyst-like that a distinct border of the unilocular radiolucency [4]. Due to tumor expansion, displacement of neighboring teeth is much more common than root resorption. The follicular variant (70.8%) [16] has been associated with unerupted teeth and scattered miliary calcifications. The manifestation resembles an odontogenic cyst and calcifying odontogenic cyst [15]. Therefore, it is easily misdiagnosed as a dentigerous cyst, ameloblastoma, or calcifying odontogenic cyst [3, 8, 18]. Clinical and radiographic manifestations of this case (follicular) extremely conform to what has been stated in the literature regarding AOT presentation [3]. The only difference in this case was that tumor appeared in the mandible, which has a lower incidence rate. The extra-follicular type (26.9%) [16] has a central lesion and no connection with the teeth, usually presenting as a unilocular well-defined radiolucency. The extraosseous type (2.3%) [16] usually presents as gingival swelling, located palatally or lingually relative to the involved tooth. All AOTs presented in this study were intraosseous: 10 (76.92%) AOTs were follicular and 3 (23.08%) AOTs were extrafollicular. There were no extraosseous/gingival AOT types in the cases collected in the article, which are associated with a low incidence of extraosseous/gingival AOT. Histologically, it not only contains the epithelium but also calcified tissue, showing a distinctive histological morphology. AOTs have a variety of

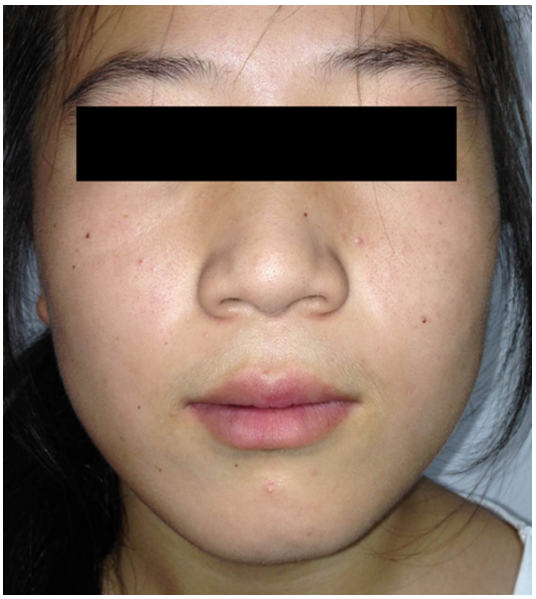

Figure 6. After decompression, the patient showed facial symmetry and the face of the patient had been greatly ameliorated.

Figure 7. Patient was followed up for 4 years, panoramic radiograph showed that bone mineral density in the diseased area was almost restored to normal bone mineral density.

characteristics, rosette-like structures consisting of cuboidal or spindle-shaped epithelial cells and glandular duct-like structures consisting of cuboidal or low columnar cells are major characteristic of AOT [1, 19]. Clinical and radiological features of AOTs might be troublesome for an inexperienced clinician. Its demanding histopathological features require careful examination from pathologists. The case presented in this study displayed rosette-like structures with minimal connective tissue.

Adenomatoid odontogenic tumors as a kind of odontogenic tumors. They have the biological nature of benign tumors, without local invasion. Recurrence rates are low. In the treatment of AOT, most authors now believe that complete enucleation with long-term observation is the preferable [1, 11, 15, 19]. For smaller AOTs, surgical enucleation and curettage of lesions can produce excellent prognosis. However, for younger patients with a larger AOT, surgical enucleation or curettage has a greater risk. The risk of damage to adjacent anatomic structures, such as the inferior alveolar canal, mandibular fracture, mandibular continuity defect, maxillary sinus, and nasal cavity, exists. Additionally, it is at the peak of growth and development for younger patients and mandibulectomy and maxillectomy with simultaneous reconstruction of the surgical defect with fibular or other flaps is inappropriate. Guided tissue regeneration with a membrane technique and bone grafting have been suggested after complete removal of the tumor [20], but long-term effects of treatment have remained unstable and controversial. The present study reviewed the relevant literature, finding that recurrence rates of AOT were relatively low in the literature and clinically. Only three cases recurred in the 750 cases reported by Philipsen and Reichart [4, 21]. Therefore, this study advocates that decompression or marsupialization can be the initial modality of treatment for younger patients with large AOTs. With the concept of minimally invasive surgery and functional surgery widely accepted, the goal of treating large cystic lesions of the jaws is to minimize postoperative recurrence and preserve the shape and function of the jaws. Moreover, fenestration decompression was introduced as a conservative treatment for odontogenic cysts, having been widely accepted. Fenestration decompression is the treatment that creates an opening to reduce pressure within a cystic cavity, inducing bone formation [12]. The rationale for fenestration decompression is to reduce the size of cystic lesions and to reduce the difficulty of total removal. Many odontogenic cysts have been treated with decompression, achieving the goal that of complete removal of the lesion and reduction of complications. After fenestration decompression, patients were required to review regularly to observe changes of tumor size and bone density [13]. When the tumor does not decrease in size, bone density is increased by about 50% [12] or continues growing during treatment by fenestration decompression, thus immediate surgical enucleation should be performed. To some extent, the treat-

ment can reduce damage to patients with large AOT. However, they will need to undergo a secondary surgery.

In the present case, fenestration decompression was chosen as treatment method for large AOT, along with enucleation as the treatment for small AOT. After the decompression of the above cases, an individual obturator was made with acrylic resin. Patients were informed to clean the obturator three times a day using saline solution to avoid obstruction and infections. Patients were scheduled for radiographic and pulp vitality test follow-ups after an interval of three months. Six months post decompression, the diminished lesion was enucleated completely under general anesthesia with an intraoral approach. No bone graft was placed in the cavity (**Figure 7**). The impacted mandibular canine was extracted. The postoperative course was uneventful and no complications occurred. There were no signs of recurrence and no apical resorption of the adjacent teeth at the follow-up after 50 months after surgery. Prognosis remains excellent.

Conclusion

Adenomatoid odontogenic tumors (AOT) are exceptionally rare and benign. They are mostly found in females. AOTs occurring in the mandible are less than those in the maxilla. However, this study showed AOT to be more common in the mandible, which may be associated with the insufficient number of cases. Conservative surgical enucleation is the treatment modality of choice for AOT. However, for large AOTs, decompression and marsupialization may be a better choice. Due to the small number of analogous cases, further studies are necessary to demonstrate the effects of decompression on large AOTs.

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Disclosure of conflict of interest

None.

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