Anterior Adenomatoid Odontogenic Tumor of the Mandible - A Case Report

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ABSTRACT Introduction: Practitioners often misdiagnose Adenomatoid Odontogenic Tumor (AOT) due to its clinical appearance that resembles dentigerous cysts and ameloblastoma. It represents hamartomatous malformation and has ductus-like structure and rose-like appearance with varying degrees of changes in its surrounding connective tissue. This tumour is of benign and non-invasive type and has low recurrence rate. It is very crucial to conduct histopathological examination as an essential adjunct in discussing clinical, radiographic and histological findings in addition to enucleation biopsy as the surgical management. Purpose: This study aims to report the management of adenomatoid odontogenic tumour found in the anterior region of mandible. The findings of this study will keep the practitioners updated in establishing the diagnosis of AOT compared to other jaw lesions. Case Report: A 15-year-old male patient consulted with chief complaints of a 3 x 2 x 1 cm asymptomatic swelling in the left anterior portion of the mandible for six months before being referred to our department. The swelling initially appeared in patient’s gum and had developed to be in more significant and more extended size. The surface of the lesion was smooth with firm & immobile consistency and associated with mandibular lateral incisors & canines impaction. Discussion: Adenomatoid Odontogenic Tumor is an uncommon benign epithelial lesion and represents only 2-7% of all odontogenic tumours associated with unerupted permanent canines. It has a marked tendency to occur in the maxilla especially in young females. In this case, we reported an uncommon finding of adenomatoid odontogenic tumour in the anterior portion of the mandible from a young man. Conclusion: The management of AOT, in this case, was conducted with enucleation biopsy and pharmacological approach. The result of post-operative evaluation was very good.

KEYWORDS Enucleation Biopsy, Adenomatoid Odontogenic Tumor

Introduction

Ghosh in 1934 was the first to describe adenomatoid odontogenic tumour (AOT) as maxillary adenomatoid.[1] Stafne described this lesion as an uncommon entity in 1948[2] using several terminologies to identify this tumour such as adenoameloblastoma, cystic complex composite odontoma, ameloblastic odontogenic tumour, and adenomatoid odontogenic tumour. In 1971, WHO adopted the term proposed by Philipsen dan Birn [3] (AOT) indicating that the lesion has been defined as “odontogenic epithelial tumour with duct-like structure and various inductive changes in the connective tissue”. The tumour is partially cystic, and in some cases, it appears as a firm lesion. This lesion is best classified as a hamartoma rather than a neoplasm.[4] Philipsen et al. established three groups of variants of AOT (follicular, extrafollicular, and peripheral). Those variants have common histological characteristics indicating the origin of the lesions from the complex system of dental lamina or the remnants. In about 96% of the cases, the tumours appear in follicular and extrafollicular types. The peripheral variant has only been found from 18 reported cases until the present time.[6] Follicular type
of AOT is mainly associated with the crown and frequently considered as the portion of impacted tooth.

Clinically and radiographically, the follicular type of AOT is often misinterpreted as dentigerous cyst.[3] We presented AOT case with cyst-like appearance found in the anterior portion of left mandible surrounding the canines that were initially misdiagnosed as dentigerous cyst.

**Figure 1:** Pre-operative : (Left) Extraoral Profile; (Right) Intraoral condition.

**Figure 2:** Pre-operative Orthopantomogram.

**Figure 3:** a. Surgical Specimen; b. Enucleation Biopsy.

**Figure 4:** Evaluation of the 14 Post-Operative Days a. Extraoral Profile; b. Intraoral; c. Gauze removal; d. Essential e. Obturator Placement.

This cyst develops from the remnants of dental epithelium tissue.[4] This cyst can expand and is often associated with cortical erosion and expansion. The expansion occurs due to the increase of osmolarity that results in the formation of inflammatory cells and epithelial desquamation into the cyst lumen. The normal distance of dental follicle is 3-4 mm. It can be suspected as dentigerous cyst if it exceeds 5 mm in diameter. The internal of the cyst has fluid accumulation containing cholesterol crystals and giant cells. [2]

Although dentigerous cyst can have relatively large size followed by a destructive bone expansion, it is an asymptomatic type of cyst.[5,9] It radiographically appears in unilocular radiolucency with some cases show multilocular appearance.[3,4]

Histopathologically, the dentigerous cyst is covered by a stratified thin layer of squamous epithelium.[8] The epithelial layer consists of 2-4 layers of flat and cuboidal cells, which are considered as the remnants of non-keratinized enamel epithelium. [12-13] Densely chronic inflammatory cells are often found in the connective tissue stroma.[8]

From all the treatment options for dentigerous cysts, enucleation is the preferable procedure. Enucleation procedure aims to remove the entire tumour mass from the wall, or its covering layer.[7] Marsupialization is usually conducted to remove large dentigerous cysts given their significant size, their ability to have destruction of surrounding tissue and the possibility of pathological fractures.[1] The interest and relevance of this case arise from the difficulty to accurately diagnose the case radiographically and histopathologically. The initial histopathological examination, in this case, revealed the findings of dentigerous cysts. However, further findings suggested that the lesion, in this case, was following the clinical characteristics of the adenomatoid odontogenic adenomatoid tumour (AOT).

**Dentigerous Cyst**

In about 24% of all the cyst cases found in jaw[1,3], the dentigerous cyst is the second most common odontogenic cyst to occur after radicular cyst.[1,2,9] Males are more affected than females, and it is mostly limited to occur in mandible.[4] Dentigerous cyst affects all types of tooth with molars and canines are the most affected teeth.[3,6] It is often found in the second to fourth decade of patient’s life.[3,5,9,11]
Both dentigerous cyst and AOT are benign encapsulated types of lesion. Enucleation procedure will help remove the lesions with no significant difficulty. If the dental follicle is not involved during the procedure and can be easily unattached from the tumour, it is possible to remove the lesion without having to extract the tooth.

In this case, the permanent canine was impacted in the large-sized tumour and its proximity to the erupted tooth made it challenging to preserve the tooth. There was no aggressive behaviour observed from the adenomatoid tumour and the recurrence following the primary lesion enucleation is rarely to occur.

Case report
A 15-year-old man consulted to the outpatient clinic of the Department of Oral and Maxillofacial Surgery, RSUP Dr Hasan Sadikin with the chief complaint of gum swelling in his left mandible. The patient initially recognised the swelling with the size of a peanut six month before being referred to our clinic. The swelling had enlarged to the size of a quail egg. The swelling mass was asymptomatic with no history of trauma. No history of weight loss and systemic disease reported.

On the extraoral examination, solitary oval swelling with a size of ± 3 x 2 x 1 cm was noted. The swelling was firm and smoothly presented with crepitation from the palpation. The inferior alveolar nerve was not affected with no pathological signs from the lymph nodes examination.

Intraoral examination revealed a solitary swelling in incisive, canine, and premolar regions of the left mandible. The buccal cortical bone expansion was noted in first premolar region of the left mandible. The surface of the swelling was smooth with a normal colour of overlying mucosa. On palpation, mild tenderness was present with partially cystic consistency as well as crepitation.

Panoramic radiograph showed destructive and expansive unilocular radiolucent lesion with a sclerotic peripheral appearance on the left mandible corpus extended to the inferior margin and slightly to the midline of right mandible.

Aspiration conducted in the right mandible, revealing clear yellowish cholesterol crystal fluid. Furthermore, the treatment of choice was enucleation biopsy as a definitive therapy to identify the type of the cyst and to establish the differential diagnosis. The biopsy result revealed that the lesion was an Adenomatoid Odontogenic Tumor (AOT).

Considering the patient’s age, the definitive treatment was done immediately to prevent recurrence possibility as well as the occurrence of massive infiltrative and destructive lesion and to avoid pathological fracture.

Intra-operatively, massive cystic lesion was obtained from the mandible corpus to left inferior margin. However, the placement of reconstruction plates was not conducted considering the density and thickness of the bone.

From the histopathological examination, the lining of AOT wall originating from dentigerous cyst epithelial remnants was obtained as the surgical specimen. Others believed that the tumour had a striking tendency of arising from the remnants of dental lamina complex system. Mature adenoid cells with normal peripheral nucleus and normal muscle tissue were noted with no signs of malignancy. The findings revealed that the tumour was best diagnosed as AOT.

The result of 7 post-operative evaluation days revealed no dehiscence as well as infection signs observed. Evaluation from the 14 postoperative days showed intraoral tissue healing. The gauze was removed, and patient was consulted to have obturator appliance (Figure 5).

Discussion
Adenomatoid Odontogenic Tumor is a non-invasive benign odontogenic lesion with slow progression of growth. It occurs in 3% of all odontogenic tumours with anterior maxillary predilection.[5] In this case, patient was a young adult man in his second decade of life presented with the lesion in his left anterior portion of the mandible.

Radiographically, this lesion was surrounding the unerupted tooth and appeared in corticated radiolucency with radiopacity. AOT often occurs covering the crown and the root of the tooth, unlike the dentigerous cyst lesion that covers the root.[3] The lesion appeared in unilocular radiolucency with well-defined cortical margin covering the crown and the root of the impacted canine.

The pathogenesis of AOT is still controversial. Some believe that the lesion is originating from the odontogenic epithelial of a dentigerous cyst.[1] Hypothetically, the theory of AOT follicle arises from the unerupted dental follicle is quite convincing.

It has been reported that most cases of AOT occur in all type of cyst and neoplasm including dentigerous cyst, calcifying odontogenic cyst, odontoma, and ameloblastoma.[7] Neoplastic and hamartomatous lesions occur in every stage of odontogenesis and as a result, odontogenic tumour with the combination of epithelial and mesenchymal features will appear.[1,8] By dentigerous cyst, AOT can show, roughly and microscopically, one or more cystic spaces covered by stratified squamous epithelium resembling the lining of dentigerous cyst as demonstrated in this case.[7]

Clinically, the radiograph and macroscopic findings, in this case, are consistent with the initial description of the lesion found in the literature. If the tumour grows following the cystic expansion, this will emphasise the origins of dentigerous cyst. However, if the development occurs before cystic expansion, the tissue will fill the follicular space and AOT appears as a solid tumour.

Radiographically, AOT should be distinguished from dentigerous cyst when observing peri-coronal radiolucency of the jaw. Dentigerous cyst only covers the crown portion of the impacted tooth. Meanwhile, the AOT shows radiolucency covering the coronal and radicular aspects of the impacted tooth. However, in the case where AOT grows from dentigerous cyst, the radiograph will show irregularities of the cyst wall revealing the development of AOT.

Only 14 cases of AOT associated with dentigerous cyst found in medical literature. From the total of the cases, nine patients were females, and five patients were males showing AOT and dentigerous cyst as benign, encapsulated, and conservative lesions.

A conservative surgical enucleation procedure is one of the modalities of the treatment option. For periodontal intra-bony deficiency caused by AOT tissue regeneration, it is suggested to undergo complete tumour removal. The recurrence of AOT is not frequent. Only 3 cases in Japanese patients reported the recurrence of AOT. [5] Therefore, the prognosis of adenomatoid odontogenic tumour is very good. No recurrence is observed from this presented case.
Conclusion

Adenomatoid Odontogenic Tumor is often misdiagnosed as dentigerous cyst observed from the radiography and histopathology examinations. Some cases of AOT occur in children. Some were also reported to have an association with the dentigerous cyst. This case highlights the importance of acknowledging the fact of unilocular lesion surrounding the impacted tooth in the anterior portion of the left mandible and establishing the proper management to have the AOT removed.

Competing interests

The authors declare no conflict of interest.

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References


