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Case Report

Mandible Adenomatoid Odontogenic Tumour Mimicking Calcifying Epithelial Odontogenic Tumour: A Rare Case Report

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Abstract

Introduction: Adenomatoid odontogenic tumour (AOT) is a rare tumour of epithelial origin predominantly found in maxilla but rarely found in mandible. Calcifying epithelial odontogenic tumour (CEOT) is a locally aggressive benign odontogenic neoplasm arising fromepithelial tissue. This case report describes a case in the mandible are where the AOT mimics CEOT. **Case presentation:** A 13-year-old female patient was referred to Persahabatan HospitalJakarta, presented with a painless lump in the lower left mandible and no exudates. Initially, the lesion was 3x2 cm in size but continued to grow to the size of a quail egg in three months. The patient came with no facial asymmetry, limited mouth opening, and tooth mobilization. **Result:** The surgical management of the lesion was enucleation and removal of associated impacted tooth. The prognosis is good and recurrences are very rare after complete removal of lesion. **Conclusion:** The combination of the partly cystic and solid tumour of AOT/CEOT proved the complexity found in odontogenic epithelial tumour cases, however, the definitive diagnosis was confirmed through its histologic features.

Keywords: Adenomatoid odontogenic tumour, Calcifying epithelial odontogenic tumour,Enucleation.

I. INTRODUCTION

Adenomatoid odontogenic tumour (AOT) is an odontogenic epithelial tumour characterized by its benign, gradually developing, and non-invasive nature. The lesion occurs in the intraosseous and in peripheral forms. AOT affects young individuals with a female predominance occurs mainly in the second decade.[1] This

tumour usually appears as an asymptomatic, slow growing swelling associated with an unerupted tooth.[1,2] It is seen more frequently in female patients and occurs most frequently during the second decade of life. Maxilla is more commonly involved and rarely in the mandible, with images impacted canines, sometimes associated with dentigerous cysts.[2] The microscopic features include the presence of ductiform structures formed by a single row of cuboidal or low columnar cells. Nodules of spindle-shaped or cuboidal cells forming sheets or rosette-like structures are found surrounding the ductiform areas. In addition, droplets of eosinophilic amorphous material, sometimes with evidence of mineralisation can be detected between the epithelial cells. A trabecular or microcystic pattern formed by round or spindle-shaped cells is found at the periphery of the lesion.[3] Radiographically, the lesion is characterized by well-defined, unilocular, radiolucent lesion associated with an impacted permanent of supernumerary tooth on the radiographic images. Therefore, the differential diagnosis of AOT varies and might be difficult to conclude, especially with other teeth-associated cystic lesions, such as calcifying cystic odontogenic tumour.[4]

Calcifying epithelial odontogenic tumour (CEOT) is an odontogenic neoplasm completely derived from the epithelial tissue.4 It has a peak incidence at around 40 years of age, with no sex or racial predilection and its main location has been the mandibular molar region.5 Microscopically this tumour is composed of sheets or strands of polyhedral epithelial cells with eosinophilic cytoplasm, well-defined cell borders and distinct intercellular bridges.[4,5] Cellular pleomorphism and prominent nucleoli are frequently found, but mitoses are rarely seen. Radiographically it appears as an ill- defined radiolucent area in which mineralised structures of varying size may be present. Approximately half of the cases have been associated with an unerupted or embedded tooth, or teeth.[5]

This case report describes a case where the AOT mimics CEOT found in the mandible area in a 13-year-old female arising in a rare location, the anterior part of mandible, with an insight into the clinical, radiographic, and microscopic manifestation of the lesion.

II. CASE REPORT

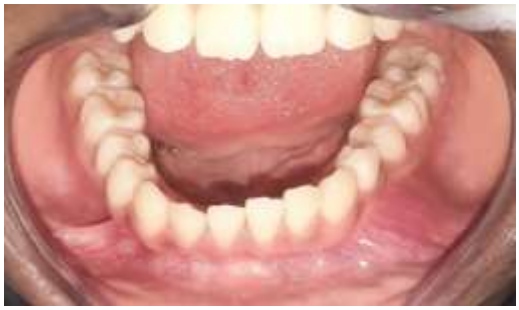
A 13-year-old female presented with a growing lump in the left area of the mandible that started to appear 3 months prior to coming to Persahabatan Hospital. The patient was referred to the hospital after receiving treatments in Wijayanti Clinic and was then referred to the Cibitung Hospital, and further referred to Persahabatan Hospital to receive medical treatment. The lump slowly grew from 2x3 cm in size to a quail egg size in three months. The lump was painless. There was no history of exudates nor salty taste inside the mouth. There was no mobility of teeth surrounding the lump, no history of dizziness, and vomiting. Patient had normal diet and body mobilization.

Medical history examination revealed that the patient had no history of hypertension, asthma, diabetes, allergy to medicine and food. The patient was cleared from having any contact with COVID-19 patients in the last 14 days. Hemodynamic result was stable, and the patient had a body weight of 41 Kg with a body height of 160 Cm.

From the extraoral examination, there was no facial asymmetry, no limitation of mouth opening, and no tenderness upon palpation of submandibular glands colli region.

Figure (1): Extraoral examination of the patient showed no facial asymmetry

From the intraoral examination, the patient had a moderate oral hygiene. The lump is located in the left side of mandible, extending from tooth 33 to tooth 36 with a size of 3 x 2 x 2 Cm. The lesion presented with same color and temperature with the surrounding tissue, had no tenderness upon palpation and had a smooth surface with hard consistency and well-defined border. No ping-pong ball phenomenon was detected.



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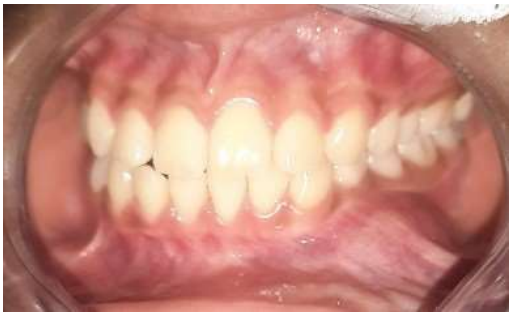

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Figure (2A, 2B): Intraoral examination of the patient showed a growing lump in left side of mandible of tooth 33 to tooth 36
Prior to the surgical treatment, the patient was referred for radiographic examination.

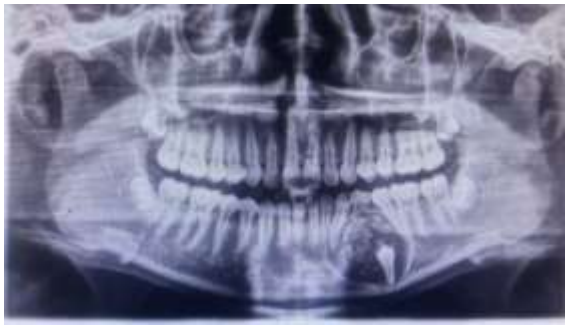
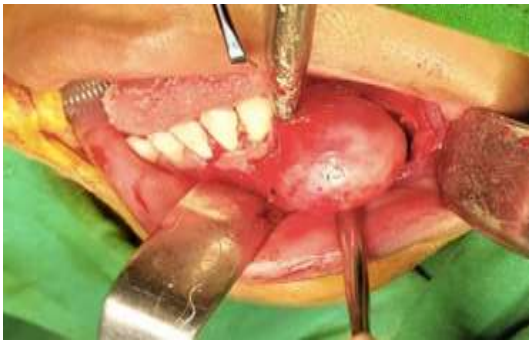


Figure (3): Radiographic examination of the patient before the surgical treatment showed a unilocular radiolucency with well-defined border and impacted tooth 34.
The treatment for this case was radical curettage and odontectomy of tooth 34 and extraction of tooth 74 under general anaesthesia.



A

B

Figure (4A, 4B): The surgical treatment for this lesion was radical curettage and odontectomy of tooth 34 and extraction of tooth 74 under general anaesthesia

Figure (5): Specimen delivered to the pathological department for histologic examination

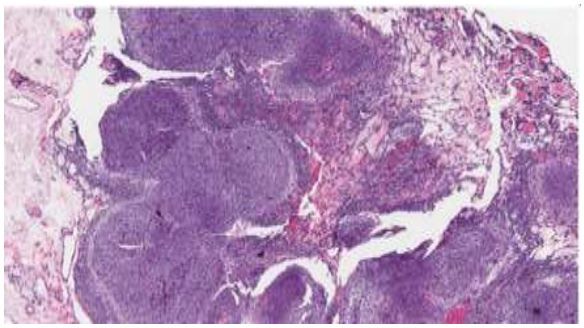


Figure (6): Histological finding of the tumour confirmed the diagnosis of adenomatoid odontogenic tumour

Histologically, the lesion showed that the specimen originated from the left mandible and showed a mass tissue of tumour with duct-like structures of the border and solid islands. Some of the peripheral cells was palisaded. The tumour cell nucleus is round/oval, showed a slight pleomorphic shape, with eosinophilic cytoplasm. There was no mitosis found. There were also calcification and eosinophilic material found. The result of the histopathological test confirmed that adenomatoid odontogenic tumour was the definitive diagnosis of the lesion. The patient was then referred for radiographic examination right after the surgical treatment.

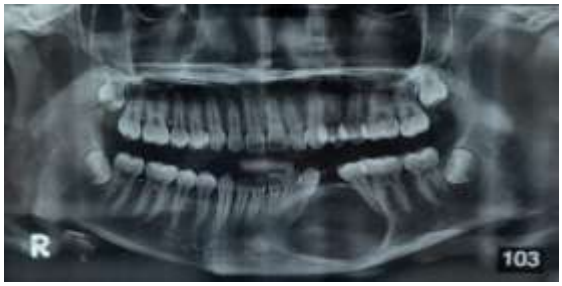


Figure (7): Radiographic examination of the patient after the treatment of radical curettage, odontectomy of tooth 34, and extraction of tooth 74 under general anaesthesia.

6 months follow-up post-surgery showed no recurrences while the patient exhibit no pain, no facial asymmetry, and no signs of growing lump in the mandible area. The patient was then taken for radiograph examination to determine the degree of bone healing.

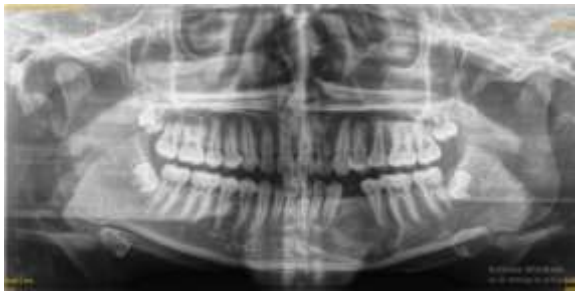


Figure (8): Follow-up radiographic examination 6 months after treatment showed bone healing and no signs of recurrences in the mandible area.

III. DISCUSSION

AOT is a slow growing, benign epithelial tumour of a nonaggressive nature.[1] It originates from the epithelial component of the tooth forming tissues, but no morphological defects were observed in the associated teeth and the progression of the tumour growth starts after odontogenesis is complete.[1,6] AOT is observed in 1% to 9% of all odontogenic tumours. Incidences are common among young females with unerupted maxillary canines.[7] This picture is consistent with this case report where the tumour occurred in a 13 year old girl, and there was a tooth that did not grow.

The present study showed that the lesion is in the canine region where AOT is mostly found, reported in previous literatures.[7] In this case report, the lesion was found in the premolars. Although the tumour displays an epithelial component, a notable pattern of typical duct-like structures, there is consistent occurrence of the tumour in a vast range of multiplicity. Foci of calcifications are usually scattered throughout the lesion. The number, size, and the degree of calcifications of these foci determine how the lesion presents radiographically.[3,7] There is a clear delineation for the AOT to occur favourably in females, ratio of female to male range from 3.2:1 in Asian and 5.6:1 in African countries. The common variant of AOT is the follicular type (70.8%), as portrayed in the present case, and extrafollicular occurring in 26.9% of the cases, peripheral type is unusual seen in 2.3 % of all AOTs. Predictable site for AOT is found in the anterior maxillary region accounting for 88.0%.[1,6]

AOT is often seen in the maxilla. However, in the present case, it was seen in the mandible. The tumour commonly is called as “two third tumour,” as two-third of the cases are seen in maxilla, two-third of the affected individuals are young female, two- third of the cases are seen to occur in association with un-erupted tooth, and two third to be affected are the canines. However, this lesion may appear as other odontogenic tumour/cyst.[1,3] However, here, the presentation was in a young female and in the mandibular and premolar region. Hence, only 2 out of the 4 features mentioned for the two-thirds tumour matched in the case presented.

Advanced stage of AOT shows swelling. As it is painless, patients delay the treatment that increases the risk of complications which includes the facial asymmetry and/or functional disorders commonly.[2,8] In the case presented, a painless well-defined lesion was seen with marginal facial assymetry. Radiograph showed unilocular lesion with tooth root involvement.

On the other hand, CEOT has been defined as a locally invasive epithelial neoplasm with a well-recognised histopathological pattern, characterised by the development of intraepithelial structures, probably of an amyloid-like nature, which may become calcified and liberated as the cells break down. Although the presence of intratumourally nodules of CEOT like epithelial cells has led some authors to consider these areas as true foci of CEOT , it is important to note that such previously reported examples, as well as those presented here, have behaved as typical AOT cases, with no recurrence or aggressiveness. [8,9] The existence of nodules of variable size, composed of polyhedral eosinophilic cells which were very similar in appearance to CEOT within our AOT cases suggest that this is not an unusual finding. Instead,

it seems to represent a frequent histological pattern of AOT.

To avoid considering it as an aggressive lesion, spindle-shaped cells in AOT appear to be morphologically and histologically similar to stratum intermedium cells of the enamel organ which, according to some authors, is also the origin of CEOT cells. This could explain the co-existence of these two embryologically related cells within the most prevalent of the two lesions (AOT), which is usually identified by its characteristic ductiform and rosette-like arrangement of cells.[2,4]

Our results suggest that those cases diagnosed as a combination of AOT-CEOT are just examples of AOT in which the spindle-shaped cells of probable stratum intermedium origin have proliferated and formed foci of cells like CEOT, but without its local aggressiveness and infiltrative growth pattern. This conclusion is supported by the fact that most of the clinical and radiographical findings, and the behaviour of all the present cases as well as that of others reported, and "combined epithelial odontogenic tumour" are basically identical to those described for classic AOT. In addition, there seems to be no reported cases of "combined epithelial odontogenic tumour" in which CEOT predominates over AOT, which may possibly prove to be a true combination of two different odontogenic tumours.[1,3,5]

The radiographic findings of AOT frequently resemble other odontogenic lesions, such as dentigerous cysts, calcifying odontogenic cysts, calcifying epithelial odontogenic tumours, keratocystic odontogenic tumour, and ameloblastoma.[10] The diagnosis of this case is AOT because of the involved impacted premolar with the presence of radiopacities.[3,10] The radiopacities in a lesion are always better compared with intraoral periapical radiographs, to a panoramic radiograph. An AOT often presents with fine radiopacities, which can be beneficial in the diagnosis.[4,10]

Sometimes a follicular AOT can resemble keratocystic odontogenic tumour and a unicystic ameloblastoma. But both these lesions are more common in the mandibular posterior regions unlike an AOT. Calcifying cystic odontogenic tumour needs to be distinguished from a follicular AOT as the location is quite similar and it may be found with an unerupted tooth.[2,10]

IV. CONCLUSION

In conclusion, combined odontogenic tumours are uncommonly found and reported in the literatures. After careful clinical and radiographic examination of the lesion, the histologic finding of the tumour specimen confirmed the diagnosis of mandible adenomatoid odontogenic tumour. The combination of the partly cystic and solid tumour of AOT/CEOT proves the complexity found among the odontogenic epithelia, however, the diagnosis can be confirmed since these tumours present with their own uniqueness in its histologic features.

Conflict of Interest:

There are no conflicts of interest..

Funding:

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