

A Rare case of massif adenomatoid odontogenic tumor in the anterior region of mandible: mimicking as dentigerous cyst

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ABSTRAK

Background. Adenomatoid Odontogenic Tumor (AOT) is a rare tumor of epithelial origin. AOT appears in three clinico-topographic variants: follicular, extrafollicular and peripheral. The AOT was predominantly found in the upper jaw, and rarely found in mandible, especially at anterior mandible. AOT is a tumor of odontogenic epithelium having duct like structures, which may be partly cystic, and in some cases the solid lesion may be present only as masses in the wall of a large cyst. The surgical management of this lesion would be enucleation along with removal of associated impacted tooth. The prognosis for both of them is good and recurrences are very rare after complete removal of the lesion. **Purpose.** It is important to define final diagnose for AOT due to mimicking with DC in clinically and radiographically finding. Biopsy is still obviously necessary to the final diagnosis. **Case.** 15-year-old female patients reported with chief complain of swelling in anterior mandible. The swelling beginning 4 years ago, gradually progressed, with no history pain, discharge and patient is complaint about loss of sensation around anterior mandible. Aspiration revealed straw colored fluid thinking in the way of DC. The provisional diagnosis of DC was given due to clinical presentation and radiographic imaging. But the biopsy examination showed AOT due to duct-like epithelial cells was being found. **Discussion.** The case report illustrates characteristic clinical and radiographic features of follicular variant of AOT mimicking a DC at unusual site that is anterior mandible. AOT is thought to arise from odontogenic epithelium and associated with the impacted tooth. Rightfully AOT is a perfect imitator of DC radiographically as well as histopathologically. It usually clinically misdiagnosed as DC as both have a unilocular, well-defined radiolucency surrounding the crown of an impacted tooth. The mass was enucleated, involved teeth were extracted, and titanium plates are used to avoid pathologist fracture. The patient had uneventful postoperative recovery. Follow up of a year has not shown any evidence of recurrence. **Conclusion.** Follicular type of OAT could confuse us with DC if the support examination just only clinically finding and radiographic examination. This case could not be definitively diagnosed on clinical and radiographic features alone. Biopsy was obviously necessary to the final diagnosis.

Keywords: adenomatoid odontogenic tumor, dentigerous cyst, enucleation, lateral incisor, mandibular anterior, titanium plate

ABSTRAK

Latar Belakang. Tumor Adenomatooid Odontogenik (TAO) adalah tumor langka yang berasal dari sel epitel. Berdasarkan gambaran klinisnya, TAO memiliki varias bentuk: berbentuk folikular, ekstra-folikular, dan periferal. Umumnya TAO ditemukan pada rahang atas, dan jarang muncul pada rahang bawah, terutama jarang pada mandibula anterior. TAO memiliki gambaran histopatologis yang khas, berupa struktur seperti ductus, yang sebagian kistik, dan pada beberapa kasus dapat berupa lesi solid pada dinding kista yang besar. Penatalaksanaan bedah pada lesi ini berupa enukleasi diikuti dengan pencabutan gigi impaksi sebagai penyebab. Prognosis pada keduanya baik, dan jarang ditemukan rekurensi kasus setelah pembedahan seluruh lesi. *Tujuan.* Penegakan diagnosis pada keduanya sangat penting untuk membedakan antara TAO dengan Kista Dentigerous (KD). *Kasus.* Anak perempuan usia 15 tahun dilaporkan dengan keluhan utama pembengkakan pada mandibula anterior. Pembengkakan berawal dari 4 tahun lalu, membesar secara bertahap, tanpa disertai keluhan sakit, dan pasien juga mengeluhkan adanya kehilangan sensasi sensoris pada bagian tersebut. Dilakukan aspirasi cairan dan didapat cairan keruh kekuningan yang menyerupai KD. Pasien didiagnosis dengan KD karena gambaran klinis dan radiografis yang menyerupai KD. Setelah dilakukan tindakan bedah, spesimen dilakukan biopsi, dan didapat gambaran TAO dengan ditemukannya sel epitel seperti duktus. *Diskusi.* Laporan kasus ini menunjukkan gambaran klinis dan radiografis dari TAO jenis folikular yang menyerupai KD, yang terjadi pada mandibula anterior yang jarang terjadi. TAO merupakan tumor odontogenik yang berasal dari sel epitel, disebabkan oleh gigi impaksi. Gambaran radiografis dan klinis dari TAO menyerupai KD. Terkadang terjadi kesalahan diagnosis dengan KD, karena keduanya unilokuler, radiolusensi terbatas tegas disekitar mahkota gigi yang impaksi. Lesi dilakukan enukleasi, termasuk pencabutan gigi insisif lateral yang impaksi, diduga sebagai penyebab, dan dilakukan pemasangan plat titanium untuk mencegah kemungkinan terjadinya fraktur patologis. Tidak ada keluhan yang berarti selama masa penyembuhan. Kontrol satu tahun tidak menunjukkan adanya tanda-tanda rekurensi. *Kesimpulan.* TAO jenis folikular dapat membuat diagnosis menjadi keliru dengan KD, bila penegakan diagnosis hanya berupa gambaran klinis dan radiografis. Tindakan biopsi merupakan modalitas utama dalam penentuan diagnosis akhir.

Kata kunci: tumor adenomatoid odontogenik, kista dentigerous, enukleasi, insisif lateral, mandibula anterior, plat titanium.

INTRODUCTION

Adenomatooid Odontogenic Tumor (AOT) is a rare tumor of epithelial origin comprising 3% of all the odontogenic tumors.¹ It was first described by Steensland in 1905. In 1907, AOT was described as pseudo-adenameloblastoma by Dreibradt in 1907. Stafne in 1948 considered AOT as a distinct entity, whereas others believed it to be a variant of ameloblastoma. In 1969, Philipsen and Birn declined this thought and suggested the name 'AOT'. In 1971, the World Health Organization (WHO) adopted the term 'AOT'. Max and Stern, in 2003, coined the name 'adenomatoid odontogenic cyst'.¹ Different terminologies such as adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantinum or teratomatous odontoma have been used before

to define the lesion which is currently known as AOT. In 1971, WHO accepted the term of AOT classified into odontogenic tumors classification 1971.²

From the early 1990s onwards 65 single cases of AOT (excluding case series of more than 10 cases) have been published. The mean age was 13.2 years (range 3 until 28 years) and the female: male ratio was 2.3 : 1. The AOT was predominantly found in the upper jaw (maxilla:mandible = 2.6 : 1). Regarding the various case series published in the literature⁽³⁾ and comparing these data with the single case reports mentioned above, it has to be reasoned that the AOT has a prevalence of odontogenic tumors between 1.2% in caucasian⁴ and 9% in black african patients.³ The tumor is most often diagnosed in the second decade of life and women are about twice as many affected than men. The AOT is over two times more located in

the maxilla than in the mandible and the anterior jaw is much more affected than the posterior area. Two-third of all cases are associated with an un-erupted tooth, and two-third affected teeth are canines.³ According to Philipsen and Reichart the AOT appears in three clinico-topographic variants: follicular, extrafollicular and peripheral. The follicular and extrafollicular variants are both intrabony and account for approximately 96% of all AOT of which 71% are of follicular type.⁴ Other references mention with nearly the same number for the follicular type (73%), extra follicular type (24%) and the peripheral type (3%).⁷

Microscopically, AOT is a tumor of odontogenic epithelium having duct like structures which may be partly cystic, and in some cases the solid lesion may be present only as masses in the wall of a large cyst.² It could explain the origin of the AOT, although is still controversial. It is thought to arise from odontogenic epithelium because it occurs in the tooth-bearing areas of the jaws, is often associated with the impacted tooth, and has various components of the enamel organ, dental lamina, reduced enamel epithelium, and/or their remnants.⁸ The hypothesis that follicular AOT arise from the reduced enamel epithelium that lines the follicles of unerupted teeth is fairly conclusive and is supported by evidence that is both morphological and immunocytochemical in nature.⁹

Dentigerous Cyst (DC) are odontogenic and arising in the jaws.¹⁰ The follicular variant of AOT is usually clinically misdiagnosed as DC as both will have a unilocular, well-defined radiolucency surrounding the crown of an impacted tooth.⁵ Both is a benign, encapsulated lesions by its own epithelial, painless, noninvasive, and slow-growing tumor that does not infiltrate the bone.¹¹

Aspiration reveals straw colored fluid which helps in differentiating DC from the solid tumor clinically and grossly follicular type of AOT sometimes extends apically beyond the cement enamel junction (CEJ)¹², while DC is attached to

the tooth at the cervical region.¹³ Cut surface of the AOT gross specimen shows a solid mass or a partly cystic space¹⁴ in contrast to cystic space of DC.

The term AOT seems to be the most appropriate for these tumors unlike the ameloblastoma, is benign and present in a very low recurrence, making it unnecessary to carry out extensive and aggressive surgery.¹⁵ The surgical management of this lesion would be enucleation along with removal of associated impacted tooth.

The prognosis for both of them is good and recurrences are very rare complete removal of the lesion. There have been some rare reports of aggressive behavior on the part of AOT. As previously mentioned, AOT are usually solid but may occasionally be cystic. Very few cases, mentioned by Tajima *et al.*, have been described that arise in association with a DC.⁹

CASE REPORT

A 15-year-old female patient reported to department of Oral and Maxillofacial Surgery Department, Hasan Sadikin General Teaching Hospital with chief complain of swelling in anterior mandible. The swelling beginning 4 years ago, formerly a marble size, but she did not find any medical treatment. The swelling gradually progressed to attain its present size in four years duration with no history pain, discharge and numbness revealed. She had no systemic disease or drug and food allergy history. Family history did not show any contribution. Patient is complaint about loss of sensation around her anterior mandible.

On extra oral examination, a solitary ovoid swelling was present on midline anterior mandible extending antero-inferiorly from her chin, lengthening her 1/3 lower face. Measuring about 14 x 10 x 10 cm in size, with smooth surface, and diffuse margins. It was not tender, firm and sessile to underlying bony structure on palpation. Regional lymph node were not palpable.



Figure 1. (a) Extra oral right profile; (b) Front extra oral profile; (c) Extra oral left profile. Swelling was present on midline anterior mandible extending antero-inferiorly from her chin, lengthening her 1/3 lower face.

On intraoral examination, there were carious lesion on right mandible first molar and left mandible second molar. The left mandible lateral incisor is not seen, suspicious un-erupted. The remain teeth from left mandible first molar until right mandible second premolar are exhibited class III mobility; many were haphazardly displaced. There was slight pain during percussion of most mandibular teeth. The lesion appeared to be intraosseous, with a considerably expanded cortex that was non-tender and pliable on both labial and lingual surfaces. A solitary swelling was present on labial and lingual vestibule of anterior mandible, extending from right second premolar to left first molar. Swelling was lobulated with clearly defined margin and overlying mucosa was intact with no ulceration.

On palpation, a solitary ovoid swelling which was firm in consistency showed no evidence of discharge on digital pressure. It was not tender with no pulsation evident. Crepitation was present on palpation. The associated teeth were found to be mobile from left mandible first molar right mandible second premolar.



Figure 2. Aspiration of intra lesion liquid and straw colored yellow turbid was aspirated.

Fine needle aspiration was carried out and straw colored yellow turbid was aspirated. Microscopic examination revealed histiocytic, mast cells; polymorph macrophages and a few foam cells were present. Laboratory examination revealed increasing of albumin, globulin, LDH and glucose.

A panoramic radiograph showed a well-circumscribed radiolucency extending from the right second premolar to left first molar, with an impacted left lateral incisors displaced to the inferior border of mandible attached below the Cemento-Enamel Junction. The lesion had caused displacement and considerable external root resorption of overlying teeth, as well as thinning, expansion, and scalloping of the inferior mandibular cortex and slightly on left. The border of mandible slightly perforated, but the lesion showed irregularly dense regions of mild opacity, consistent with soft tissue proliferations, along the lesional periphery and occasionally more centrally; some of these regions had enough density to suggest at least mild calcification. The bulk of the lesion was represented by a mildly and diffusely gray haze, suggesting a fluid content. There were several small, more radiopaque densities of the central portions, and the overlying cortex was very thin, with occasional perforations not evident on other radiographs. The lesion considerably expanded toward the labial, but also showed expansion of the lingual aspect of the mandible. The central lesion shows irregular density with mixed of radiolucent and translucent portion. Radiolucent portion shows perforated lesion on lingual/buccal and inferior aspect. And translucent portion shows there was still adequate bone to keep the mandible shape.



Figure 3. A panoramic radiograph showed a well-circumscribed radiolucency, with an impacted left lateral incisors displaced to the inferior border of mandible .

The provisional diagnosis of DC was given. The mass was enucleated in toto and the cyst separated easily from the adjoining bone and was removed with the involved tooth (left mandible first premolar until right mandible first premolar). Radix right mandible first molar was extracted due to pathologist condition. 12-holes titanium plate with 3 screws, two screws on right side and one screw in left side are used. The titanium plate was placed too high until tooth furcation. But it was not a problem since the placement plate just for temporary, and no bone expose while follow up.

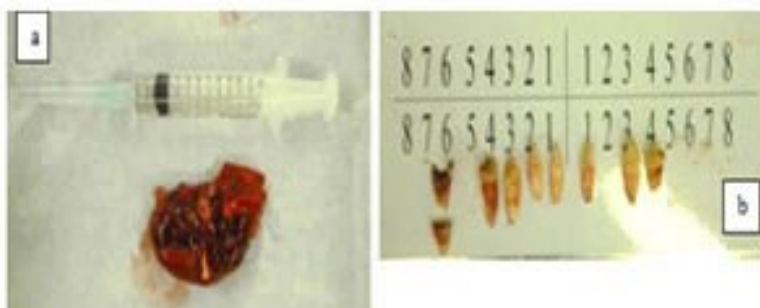


Figure 4. (a) White to brown in color gross specimen with diameter 1.3 cm in size that sent to pathologist for biopsy examination. We could see lateral incisor inside the lesion was being extracted too; (b) Multiple teeth was extracted since AOT make root resorption and tooth mobility.

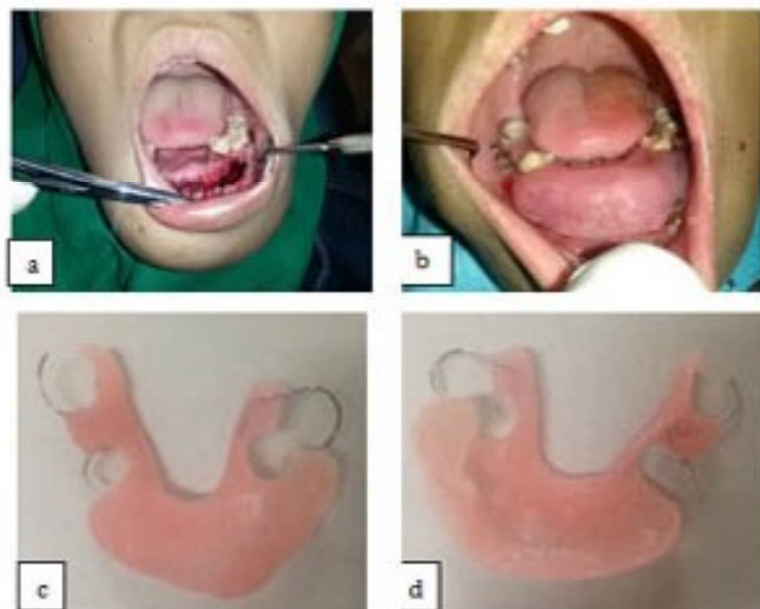


Figure 5. (a) Intra oral post-operative day 7, when obturator was molded and ready to place; (b) Obturator was placed to cover entire cavity; (c) Upper surface of obturator have been polished to reduce plaque accumulation due to smooth surface; (d) Lower surface of obturator that slightly contact with the cavity.

The gross specimen received was white to brown in color, partly solid and brittle, with a tooth inside pathologic tissue was sent for biopsy examination. It also showed a cystic space with

solid mass wall white to brown in color, 0.2-1 cm in size. Microscopically, there was ovoid to spindle cells, grows hyperplastic, dense in solid with glandular tissue, partly cribriform. The solid

portions of the tumor showed strands, large sheets, and islands of spindle-shaped, polygonal or duct-like epithelial cells, sometimes forming whorled masses of cells. Nuclear cells shows within normal limit.

Fibrocollagen stroma tissue was degraded into mixoid and hyaline. It showed lymphocyte cell with enlarge of blood vessel and area of hemorrhage. Classification was shown. There was no sign of malignancy. The histopathological diagnosis of cystic adenomatoid odontogenic tumour was given based on the above features.

The patient had uneventful postoperative recovery. Acrylic obturator was molded with lower surface of obturator slightly contact with the cavity. Every two weeks it was being reduced, to permit the bone grew from the bottom of the cavity and to prohibit the cavity closure by itself. This could prevent the recurrent of the disease too. Follow up of 1 year has not shown any evidence of recurrence. Bone formation was shown and titanium plate could be removed.

DISCUSSION

The case report illustrates characteristic clinical and radiographic features of follicular variant of AOT mimicking a DC at unusual site that is anterior mandible. Anterior mandible is rare site than other site which predominantly found in the upper jaw (maxilla : mandible = 2.6 : 1).

The origin of the AOT is controversial. It is thought to arise from odontogenic epithelium because it occurs in the tooth-bearing areas of the jaws, is often associated with the impacted tooth, and has various components of the enamel organ, dental lamina, reduced enamel epithelium, and/ or their remnants.⁸

Philipsen *et al.* also postulated that the follicular type of AOT develops from nests of cells within the dental lamina and, therefore, as a result, surrounds the tooth.⁹ They surround the crowns and are attached to the necks of unerupted teeth in a true follicular relationship. Whether origin of the follicular variant occurs before or after cystic expansion has taken place is open to conjecture. If it occurs after cystic expansion, then this effectively means origin from DC, and several such case reports have been published.⁹ If it occurs before cystic expansion, then the

tumor tissue will fill the follicular space and the AOT will present as a solid tumor. It is reasonable to assume that, given enough time, even those originating from a cyst may grow and fill the lumen completely. It cannot be ruled out that the DC with an impacted lateral incisors developed first followed by development of AOT in the cyst wall.

Gadewar and Srikant in their recent publication have kindled the controversy of Adenomatoid Odontogenic Tumour being a cyst or a tumour. Right fully AOT is a perfect imitator of DC radiographically as well as histopathologically.¹⁶ The follicular variant of AOT is usually clinically misdiagnosed as DC as both will have a unilocular, well-defined radiolucency surrounding the crown of an impacted tooth.⁵ Aspiration revealed straw colored fluid thinking in the way of DC, but on gross examination the soft tissue extended beyond the CEJ with a positive finding towards AOT, but the cut section showed both the solid and cystic areas which made us to consider a Cystic variant of Adenomatoid Odontogenic Tumor.

Histopathological features of OAT usually shows a well-defined tumor surrounded by a fibrous capsule (Fig. 6A). The tumor includes two major epithelial cell types. The first type is characterized by spindle-shaped epithelial cells arranged in sheets, strands, or whorled nodules within a scant fibrous stroma. These epithelial cells occasionally may form rosette-like structures, which surround an empty space or a small amount of amorphous eosinophilic material. This material may stain for amyloid. The second epithelial cell type is cuboidal to columnar and forms tubular or duct-like structures (hence, the term, adenomatoid) (Fig. 6B). Such tubular or duct-like structures may vary considerably in size and are not always present. They are lined by a single row of cuboidal to columnar epithelial cells. The nuclei of these cells are polarized away from the lumen. Most lesions also produce varying amounts of calcified material. There may be small basophilic calcifications, which likely represent dystrophic enamel calcifications. Sometimes these calcifications may exhibit a concentric ring pattern. In some cases, there may be larger masses of matrix or calcification. This material has been interpreted as dentinoid, osteodentin, or cementum. Dentinoid seems to result from a metaplastic process, because there is no

odontogenic ectomesenchyme present to induce dentin production. A few cases of adenomatoid odontogenic tumor with focal calcifying epithelial odontogenic tumor-like areas (referred to as combined epithelial odontogenic tumor) have been reported.¹⁶ The presence of such foci does not seem to influence biologic behavior; thus, these lesions may be regarded as a histopathologic variant of adenomatoid odontogenic tumor.

As a mimicking ok AOT, clinical and radiographic feature, microscopic features of DC usually lined by a thin layer of nonkeratinizing epithelium that is 2 to 4 cells thick (Fig. 7). The interface between the epithelium and cyst wall

is flat and devoid of rete ridges. The cyst wall is composed of loose to moderately dense fibrous connective tissue, which often includes scattered, small odontogenic epithelial rests that represent remnants of the dental lamina epithelium. Sometimes, these rests undergo dystrophic calcification. Although the dentigerous cyst is considered developmental in nature, many contain a mild to sometimes heavy chronic inflammatory cell infiltrate in the cyst wall—especially those cases that are associated with a partially erupted tooth. In such instances, the epithelial lining often demonstrates irregular hyperplasia of rete ridges (Fig. 7).

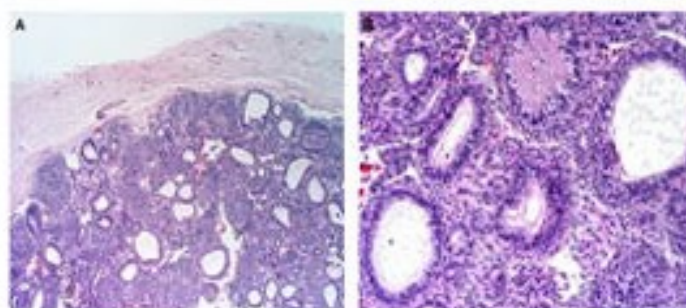


Figure. 6. Adenomatoid odontogenic tumor. (A) Low-power photomicrograph shows a well-defined, encapsulated tumor mass with a whorled growth pattern and scattered duct-like structures. (B) High-power photomicrograph shows variably sized duct-like structures and focal rosette formation within a background of spindle cells. The cuboidal to columnar cells lining the duct-like structures have nuclei polarized away from the lumens.

The epithelial lining may also include scattered mucin-producing cells, which are indicative of the pluripotentiality of the odontogenic epithelium (Fig. 7).¹⁶ In rare instances, cilia and sebaceous glands have even been reported. Finding mucous cells in a dentigerous cyst lining is not thought clinically significant, although some

investigators have pointed out that the ability to undergo glandular differentiation suggests that odontogenic epithelium may be the source of rare cases of intraosseous mucoepidermoid carcinomas of the jaws. Occasional dentigerous cysts exhibit irregular linear and polygonal calcifications within the epithelial lining (Rushton bodies) (Fig. 7).¹⁶

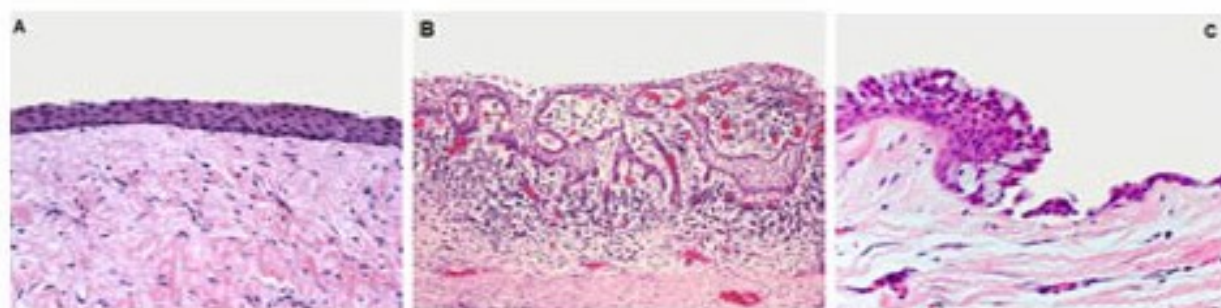


Figure. 7. (a) Dentigerous cyst. The cyst is lined by a thin layer of stratified squamous epithelium. (b) Dentigerous cyst. The presence of inflammation may result in irregular hyperplasia of the epithelial lining. (c) Dentigerous cyst. Scattered mucin-producing cells are present within the epithelial lining.

The size of the present AOT, however, is probably its most unique feature. This entity is generally perceived as one of the most innocuous of odontogenic tumors; most cases are less than 1.5 cm in diameter. In fact, the present case represents 1 of the 2 or 3 largest AOT ever reported, and it appears also to be one of the most aggressive, if not the most aggressive. It drastically resorbed virtually every root coming into contact with it. It dramatically moved and mobilized almost all overlying teeth and, in fact, moved and resorbed teeth much more than any other reported AOT. Moreover, it was so large, and expanded so rapidly, that its pressure on both mental nerves produced bilateral paresthesia. Its expanded cortex was so thin as to be almost translucent at surgery, and it was perforated in several places. No other case could be found of an AOT that perforated the overlying cortex or expanded it so rapidly that paresthesia developed.

The whole mass was enucleated and the cyst separated easily from the adjoining bone, remain massive defect. The involved teeth were extracted. And to keep the mandible sturdy, titanium plate was used to strength the mandible continuity. Acrylic obturator was molded over the cavity to prohibit the cavity closure by itself. Every two weeks it was being reduced, to permit the bone grew from the bottom of the cavity. Bone regeneration is allowed due to remaining bone at lingual site. Slow growing of bone are seen, six month later. After a year, titanium plate could be remove.

CONCLUSION

Follicular type of OAT could confuse us with DC if the support examination just only clinically finding and radiographic examination. This case could not be definitively diagnosed on clinical and radiographic features alone. Biopsy was obviously necessary to the final diagnosis. While histopathologic examination shows OAT have duct like structures which may be partly cystic, and in some cases the solid lesion may be present only as masses in the wall of a large cyst. It is most interesting, and challenging, for its abundance of "odd" features, which made the formation of a good differential or working diagnosis extremely complicated. With the perfect imitator condition,

some writer published follicular variant of OAT is originated from DC, whether origin of the follicular variant occurs before or after cystic expansion has taken place is open to conjecture.

Post-operative defect due to massive size of lesion will leave an esthetical problem, especially in young adolescence female. This problem should take a special consideration in treatment planing. Additionally, the case provides an excellent example of the extremes that can occur in even innocuous odontogenic lesions; not all cases fall within the textbook parameters.

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