

ADENOMATOID ODONTOGENIC TUMOR EXPANDING THE MAXILLA AND SHIFTING THE DENTAL MIDLINE: A CASE REPORT



Hesham Saleh Khalil*, PhD
Ahmed Zahrani*, PhD

Adenomatoid odontogenic tumor (AOT) is a benign, non-invasive lesion with slow but progressive growth. Three variants of AOT were reported in the literature: follicular, extrafollicular, and peripheral. This report illustrates an unusual case of AOT causing shifting of the dental midline. Orthopantomogram and CT Scan radiographs revealed a well circumscribed unilocular radiolucency located in the area of the left maxillary lateral incisor, canine and first premolar. The radiolucency was not associated with an impacted tooth or any calcification. Biopsy was performed and microscopic examination revealed the presence of an AOT. The patient has been followed-up for one year without recurrence.

KEY WORDS: Adenomatoid, jaw swellings, odontogenic, tumor.

J Pak Dent Assoc. 2009; 18(4): 185 - 188.

INTRODUCTION

Adenomatoid Odontogenic Tumor (AOT) is a relatively uncommon odontogenic tumor. It has a prevalence of 1.2% in Caucasian and 9% in African patients.¹ The tumor is usually associated with unerupted teeth and seldom causes root resorption. We present a case of AOT causing labial displacement of the left maxillary canine and shifting the dental midline 4mm to the right side of a 26-year old male patient.

CASE REPORT

A 26-year old male presented with a painless slow growing swelling at the left side of the upper jaw of 6 months. On examination, the swelling was 2x2cm, circumscribed, bony, hard and non tender located between the lateral incisor and the first premolar. The canine was pushed labially and rotated and the soft tissue covering the

swelling was normal in colour with the exception of an ulcerated area due to a trauma from the opposing canine. There was no history of trauma, pain or discharge and the patient was medically fit (Figure 1 a-b). The midline was shifted to the right by 4 mm. Aspiration revealed only blood. A periapical radiograph and CT Scan showed a unilocular radiolucency with no impacted teeth and no resorption of the adjacent roots (Figure 2 a-b). The lesion was completely enucleated and the compromised adjacent canine was also removed (Figure 3 a-b). The lesion was sent to the Maxillofacial Pathology Department for histological preparation and interpretation.

Radiological findings

Periapical, OPG radiograph and CT Scan showed a well circumscribed unilocular radiolucency located in the area of the left maxillary lateral incisor, canine and first premolar (Figure 2 a-b). The radiolucency was not associated with an impacted tooth and the calcified deposits which are seen in 78% of AOT² were not present in this case. No resorption of the adjacent roots was seen.

* Department of Oral and Maxillofacial Surgery, College of Dentistry, King Saud University, Riyadh, Saudi Arabia.

Correspondence: "Dr. Hesham Saleh Khalil" <hkhalil@ksu.edu.sa>



Figure 1(a & b). Photographs (a& b) showing a 2x2 cm circumscribed bony hard and non tender located between the lateral incisor and the first premolar. The canine was pushed labially and rotated and the soft tissue covering the swelling was normal in color with the exception of an ulcerated area due to a trauma from the opposing canine.

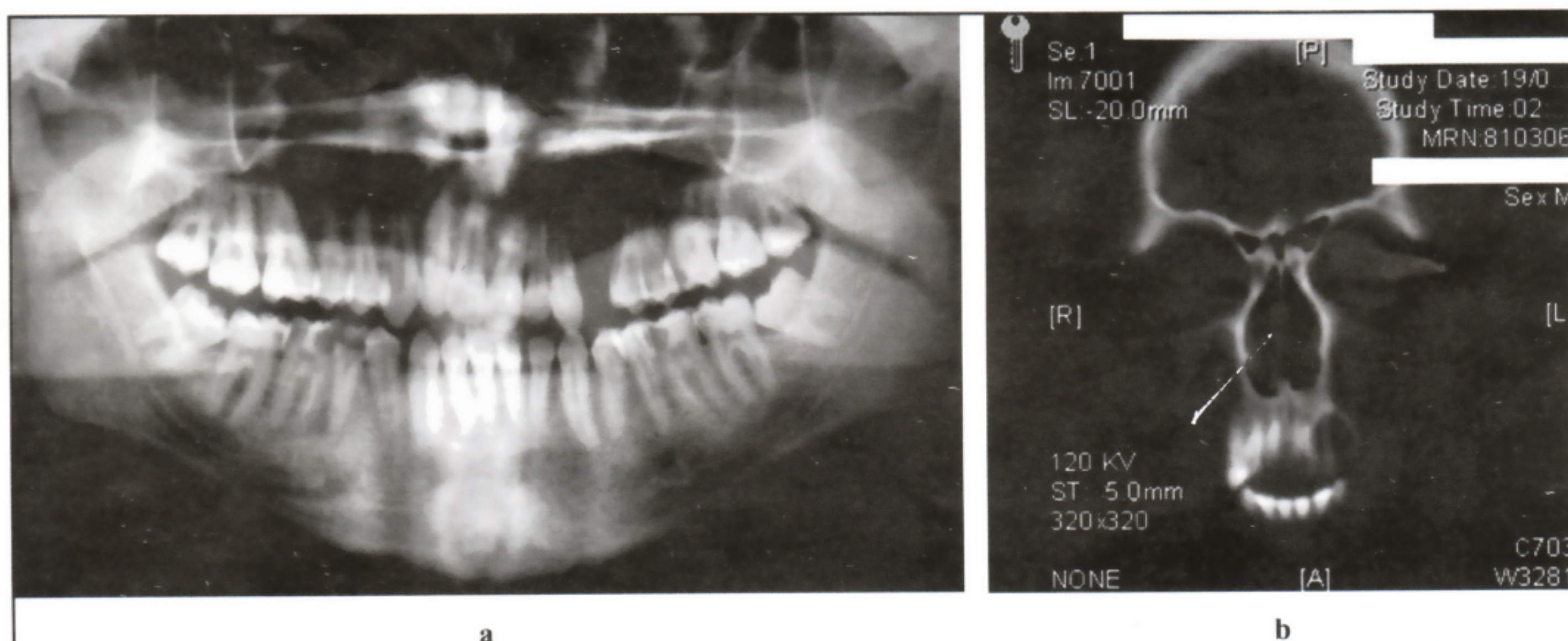


Figure 2 (a & b). Orthopantomogram (a) and coronal CT Scan (b) showing a well circumscribed unilocular radiolucency located in the area of the left maxillary lateral incisor, canine and first premolar. The radiolucency is not associated with impacted tooth and the calcified.

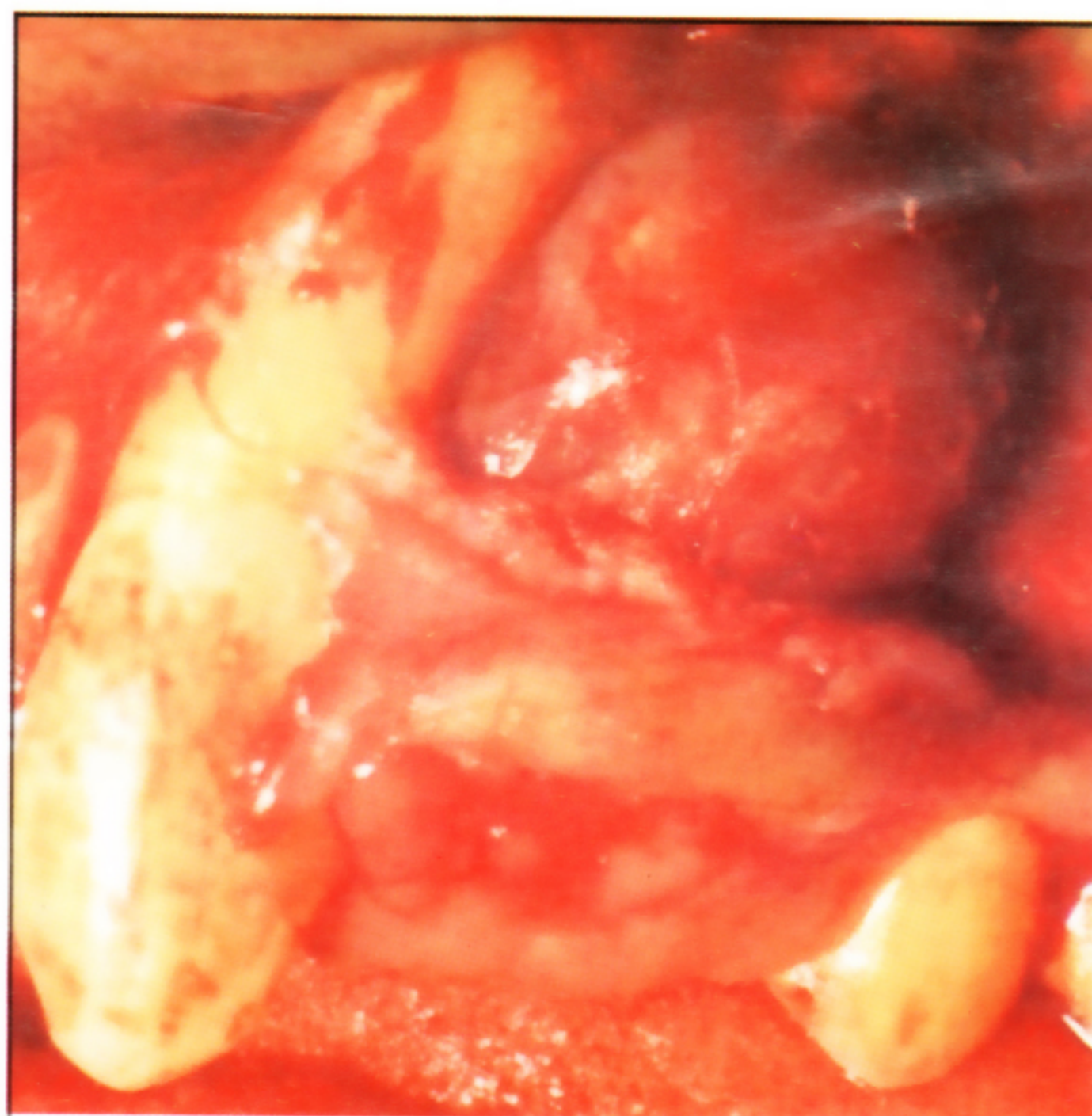
Histological findings

Histological examination revealed a capsulated fibro-vascular hyperemic mass containing sheets, stands and whorls of odontogenic epithelium (Figure 4). Rosettes and duct-like structures containing fine homogenous eosinophilic secretions were also noted. In addition, dentinoid materials and dystrophic calcification were observed in the stroma.

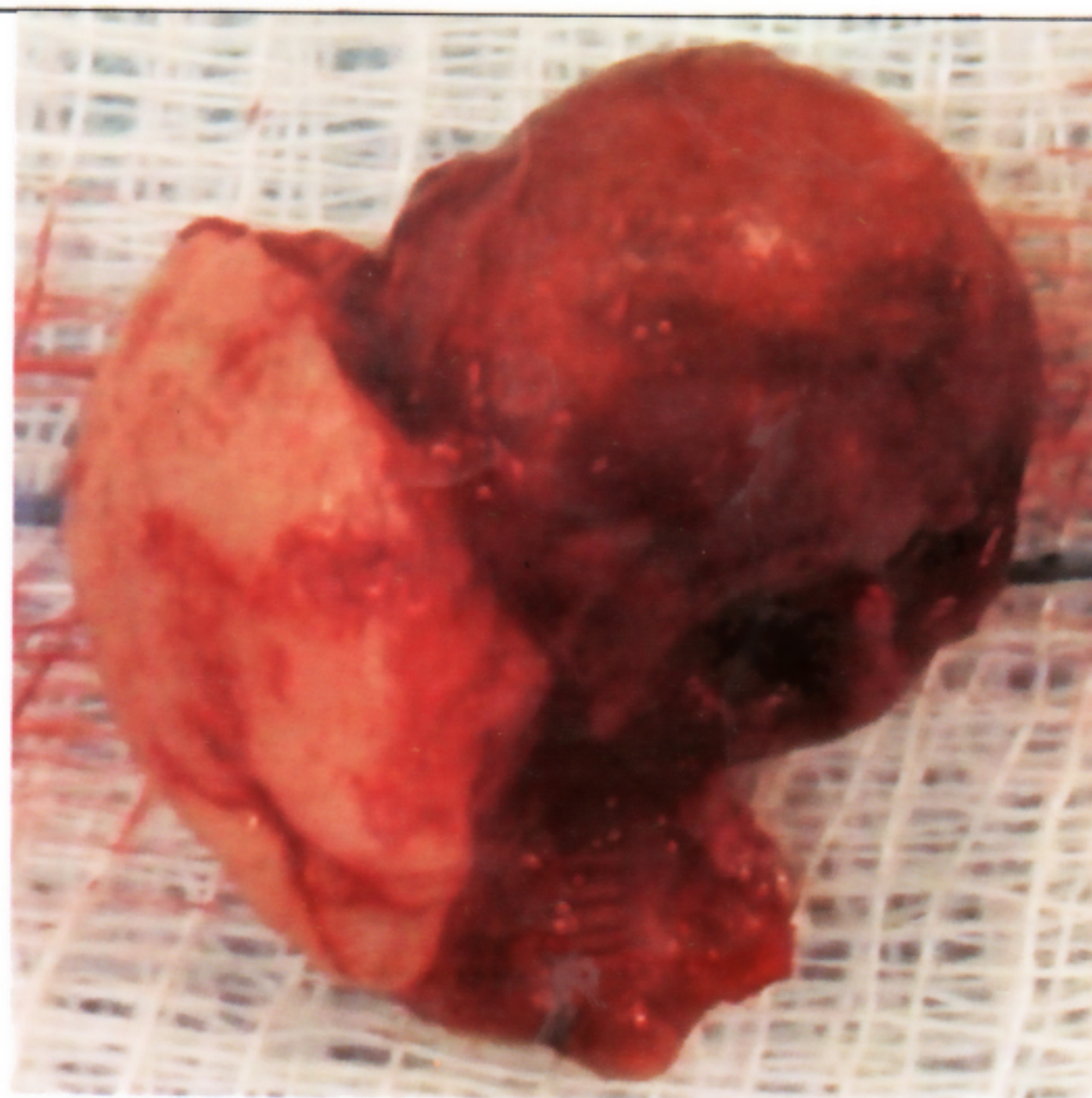
DISCUSSION

The Adenomatoid Odontogenic Tumor is an

uncommon benign tumor which makes up approximately 3 % of all odontogenic tumors.³ It occurs mainly in females in their second decade with a female to male ratio of 2.3:1.¹ AOT is mainly found in the maxilla at the lateral incisor/canine region but has been reported in different sites such as maxillary premolar area⁴, lower canine¹ and mandibular third molar region.⁵ Three types of AOT were described based on their clinicopathologic features. The follicular intraosseus lesion with associated unerupted tooth, the extrafollicular intraosseus with no associated unerupted tooth and the peripheral type which appears as



a



b

Figure 3 (a & b). Photographs showing AOT intra-operatively (a) and after complete excision (b)

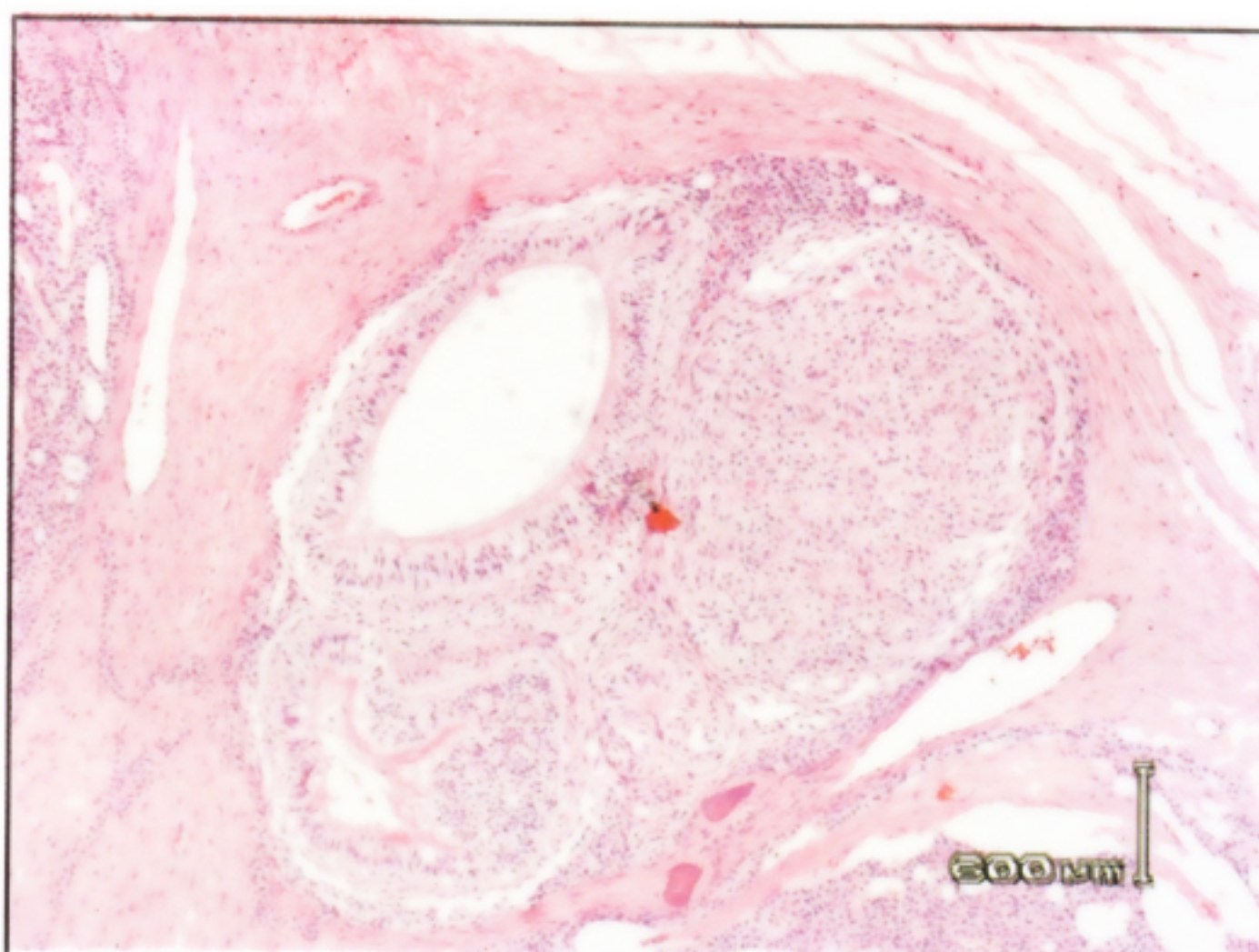


Figure 4. A histological section of the excised lesion showing a capsulated fibro-vascular hyperemic mass containing sheets, strands and whorls of odontogenic epithelium. Rosettes and duct like structures containing fine homogenous eosinophilic secretions are also noted. In addition dentinoid materials and dystrophic calcification are observed in the stroma.

gingival fibroma or an epulis attached to the gingiva.⁶

Clinically, AOT presents as a slow growing painless mass with the maxilla being affected twice as the mandible AOT is an uncommon cause of jaw swelling⁶, and it is unlikely that simple bone cysts produce root

divergence and teeth displacement, although about 20% of simple bone cysts have in fact clinically noticeable expansion.⁷ In this case report, the tumor displaced the left maxillary canine labially and generated a large gap between the canine and the first premolar. Additionally the dental midline was shifted to the right by 4 mm and the maxillary central incisors were inclined. Although in about 75% of cases AOT is associated with an impacted tooth, most often a canine,⁸ in our reported case there is no impacted tooth associated with the lesion.

AOT rarely causes root resorption and has a low recurrence rate. Surgical enucleation or curettage has proven to be the treatment of choice for all types of AOTs. In the case reported, the lesion was surgically enucleated with no signs of recurrence one year after surgery (Figure 5 a-b).

In regards to the genetics role in the formation of odontogenic tumors, it has been reported that mutations of the ameloblastin gene have been detected in Adenomatoid Odontogenic Tumor and, and ameloblastin-mutant mice develop odontogenic tumors of odontogenic epithelium origin.⁹

CONCLUSION

It is concluded that AOT should be considered in the differential diagnosis of swellings causing severe displacement of teeth.

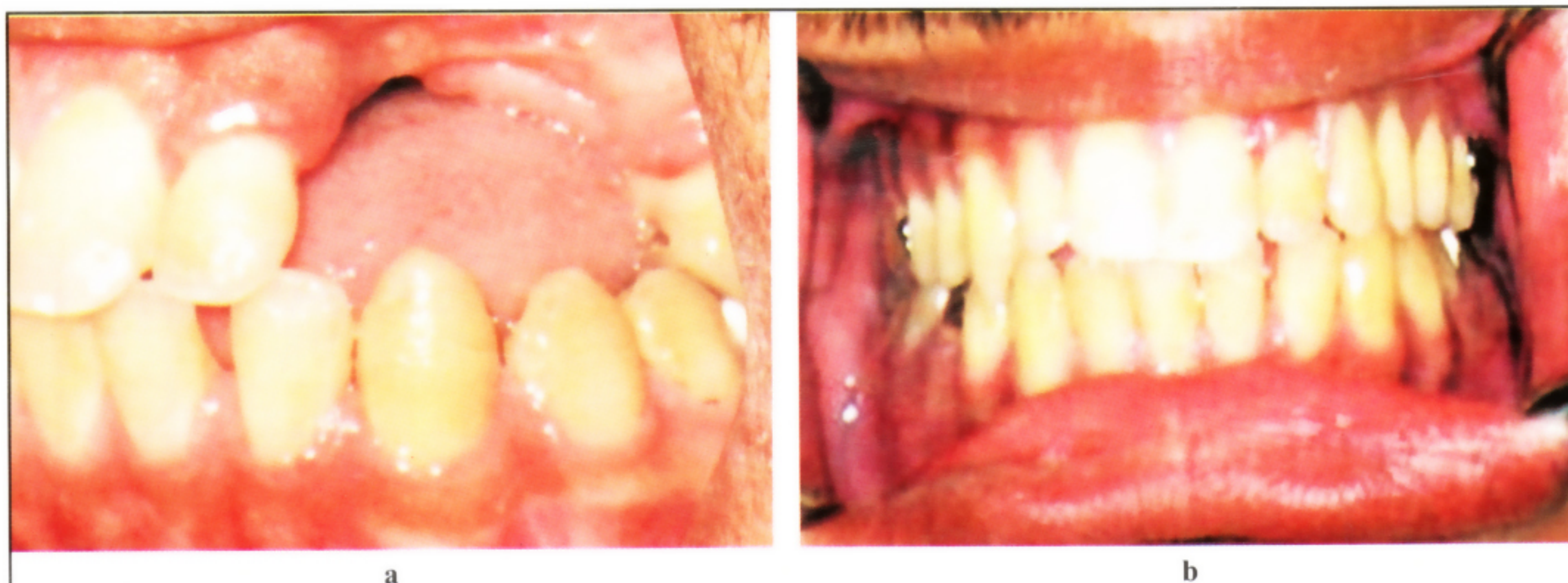


Figure 5 (a & b). Photographs showing healing 11 months after excision (a) and a removable partial denture replacing the missing teeth (b).

REFERENCES

1. Handschel, JG, Depprich, RA, Zimmermann, AC, Braunstein, S, and Kubler, NR. Adenomatoid odontogenic tumor of the mandible: review of the literature and report of a rare case. *Head Face Med*, 2005, 1:3.
2. Toida, M, Hyodo, I, Okuda, T, and Tatematsu, N. Adenomatoid odontogenic tumor: report of two cases and survey of 126 cases in Japan. *J Oral Maxillofac Surg*, 1990, 48:404-408.
3. Vera Sempere, FJ, Artes Martinez, MJ, Vera, SB, and Bonet, MJ. Follicular adenomatoid odontogenic tumor: immunohistochemical study. *Med Oral Patol Oral Cir Bucal*, 2006; 11: E305-E308.
4. Bulut, E, Tasar, F, Akkocaoglu, M, and Ruacan, S. An adenomatoid odontogenic tumor with unusual clinical features. *J Oral Sci*, 2001, 43: 283-286.
5. Sato, D, Matsuzaka, K, Yama, M, Kakizawa, T, and Inoue, T. Adenomatoid odontogenic tumor arising from the mandibular molar region: a case report and review of the literature. *Bull Tokyo Dent Coll*, 2004, 45: 223-227.
6. Nigam, S, Gupta, SK, and Chaturvedi, KU. Adenomatoid odontogenic tumor--a rare cause of jaw swelling. *Braz Dent J*, 2005, 16: 251-253.
7. Hisatomi, M, Asaumi, J, Konouchi, H, Yanagi, Y, Matsuzaki, H, and Kishi, K. Comparison of radiographic and MRI features of a root-diverging odontogenic myxoma, with discussion of the differential diagnosis of lesions likely to move roots. *Oral Dis*, 2003, 9: 152-157.
8. Fregnani, ER, Pires, FR, Quezada, RD, Shih, I, Vargas, PA, and de Almeida, OP. Calcifying odontogenic cyst: clinicopathological features and immunohistochemical profile of 10 cases. *J Oral Pathol Med*, 2003, 32: 163-170.
9. Perdigao, PF, Gomez, RS, Pimenta, FJ, and DE Marco, L. Ameloblastin gene (AMBN) mutations associated with epithelial odontogenic tumors. *Oral Oncol*, 2004, 40: 841-846.