

UNICYSTIC AMELOBLASTOMA: RAISING OBSCURITY  
IN PERIRADICULAR DIAGNOSIS - A Case Report

## ABSTRACT:

Ameloblastoma is a benign tumor; whose importance lies in its potential to grow to enormous size with resulting bone deformity. Unicystic ameloblastoma (UA) is a less encountered variant of ameloblastoma. The neoplastic nature of the lesion becomes evident only when the enucleated material is available for histologic examination. Although its predilection is for the posterior mandibular region, but its presence in the periradicular area mimicing a cyst and a granuloma cannot be overlooked upon. Despite the clinical diagnosis of periapical disease of endodontic origin, a non endodontic lesion may be present. Hence, lesions such as UA located on periapical area of a tooth can lead to a pulp-periapical misdiagnosis, and should be considered in differential diagnosis. This paper illustrates a case of unicystic ameloblastoma present in the anterior maxillary region that clinically and radiographically resembled an inflammatory pathosis, emphasizing the importance of correct diagnosis and treatment.

KEYWORDS: Unicystic ameloblastoma, differential diagnosis, periapical lesion.

Date of Submission : 11/1/16

Date of Acceptance : 21/2/16

1. Ramandeep S Narang
2. Adesh S Manchanda
3. Jaspreet Kaur

1. MDS, Professor, and Head Department of Oral Pathology & Microbiology, Sri Guru Ram Das Institute of Dental Sciences and Research, Amritsar.
2. MDS, Reader, Department of Oral Pathology & Microbiology, Sri Guru Ram Das Institute of Dental Sciences and Research, Amritsar.
3. BDS, Sri Guru Ram Das Institute of Dental Sciences and Research, Amritsar.

Corresponding author:

Name: Dr. Adesh S Manchanda

MDS, Reader, Department of Oral Pathology & Microbiology,

Sri Guru Ram Das Institute of Dental Sciences and Research, Amritsar.

e-mail: adesh\_manchanda@yahoo.com

(M) : 07837060067

## INTRODUCTION

Odontogenic tumours (OTs) are derived from epithelial or mesenchymal elements, or both, that are part of the tooth forming apparatus. They are therefore exclusively found in the mandible and maxilla and must be considered in differential diagnosis of lesions involving these sites. Like neoplasms elsewhere in the body, OTs tend to mimic microscopically, the cell or tissue of origin. An understanding about the biologic behaviour of the various OTs is of fundamental importance to the overall management of patients.<sup>1</sup>

Ameloblastoma is a benign tumor; whose importance lies in

its potential to grow to enormous size with resulting bone deformity.<sup>2</sup> Unicystic ameloblastoma (UA) is a less encountered variant of ameloblastoma and appears more frequently in the second or third decade with no sexual or racial predilection. Much confusion still exists when it comes to the terminology used for UA. Some of the terms used for this lesion prior to 1977, when Robinson and Martinez' introduced the concept of UA, were cystic (intracystic) ameloblastoma, ameloblastoma associated with dentigerous cyst, cystogenic ameloblastoma, extensive dentigerous cyst with intracystic ameloblastic papilloma, mural ameloblastoma, dentigerous cyst with ameloblastomatous proliferation, and ameloblastoma

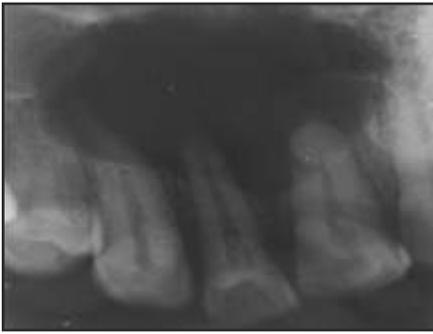


Figure 1: Intraoral periapical radiograph showing a unilocular radiolucent lesion involving the #21 tooth region to #24 tooth region.

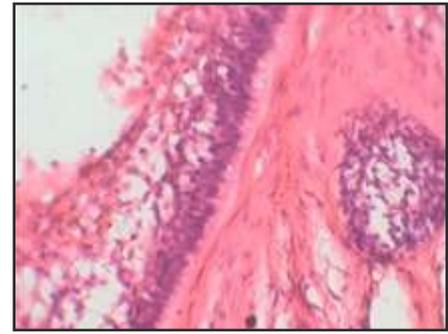
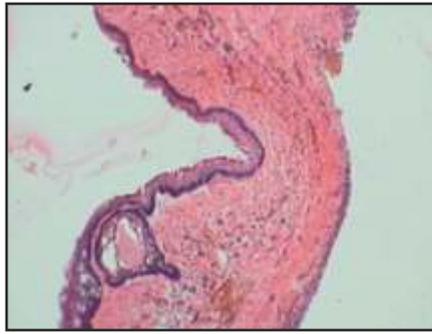


Figure 2: (a) Ameloblastic epithelial lining with palisaded basal cell layer and stellate reticulum like cells (H & E; X10). (b) Ameloblastic epithelial lining showing nuclear palisading, hyperchromatic nuclei with polarization and cytoplasmic vacuolation with intercellular spacing and stellate reticulum like cells (H & E; X40).

developing in a radicular (or "globulomaxillary") cyst. The term *unicystic* is derived from the macro and microscopic appearance, the lesion being essentially a well-defined, often large monocystic cavity with a lining, focally but rarely entirely composed of odontogenic (ameloblastomatous) epithelium.<sup>3</sup>

The diagnosis of unicystic ameloblastoma is based on two features. First, the lesion must be unilocular (less commonly multilocular), clinically and radiologically. Second, on microscopic examination it must appear as a single cystic lesion with the epithelial lining consisting of ameloblastoma. If the lesion is small, it is commonly seen as an incidental finding on radiographs taken for other purposes, in these circumstances, some lesions could remain undiagnosed in the early stages of their development.<sup>4</sup>

UA is almost exclusively encountered asymptotically in the posterior mandible, and its presence in the periradicular area mimicking a cyst and a granuloma cannot be overlooked upon. Despite the clinical diagnosis of periapical disease of endodontic origin, a non endodontic lesion may be present. Hence, lesions such as UA located on periapical area of a tooth can lead to a pulp-periapical misdiagnosis, and should be considered in differential diagnosis. The neoplastic nature of the lesion becomes evident only when the enucleated material is available for histologic examination.

This paper illustrates a case of unicystic ameloblastoma present in the anterior maxillary region that clinically and radiographically resembled an inflammatory pathosis, emphasizing the importance of correct diagnosis and treatment.

#### CASE REPORT

A 28 year old female patient presented with a swelling in the upper left anterior region of maxilla. On extraoral examination, a single, non tender swelling about 2 x 2 cm in size was observed on the left side of face. Intraoral examination revealed a soft, fluctuant swelling of size 2 x 2

cm, extending up to #11 tooth region medially and distally till #24. Hard tissue examination revealed caries present in relation to #21 and #22. On radiographic examination, a unilocular radiolucency of 2.5 x 2 cm in size was seen periapically in relation to #21 extending till #24 along with the widening of periodontal ligament space of #21, #22 and #23 (Figure 1). Aspiration cytology revealed few squamous cells in a background of inflammatory cells.

Based on the clinical and radiographic examination a provisional diagnosis of radicular cyst was given. In this case the focus was on radicular cyst which is a highly prevalent periapical lesion associated with carious tooth, pulp necrosis, and infection. Other non inflammatory periapical lesions such as periapical cemento- osseous dysplasia, bone cyst, odontogenic keratocyst, fibro-osseous neoplasm, adenomatoid odontogenic tumor and ameloblastoma were also considered in the differential diagnosis at a later stage. Surgical enucleation of the lesion under local anesthesia with adrenaline was performed. A full thickness flap was raised, a window was made in the alveolar bone, the access was enlarged, and the entire lesion was enucleated. The excised specimen was then submitted for histopathological examination.

Histologically, the given section showed a cyst wall lined by ameloblastic epithelium which was disrupted in places. The lining showed columnar basal cells with hyperchromatic nuclei; nuclear palisading with polarization and cytoplasmic vacuolation with intercellular spacing; and a thin layer of stellate reticulum like cells. The stroma showed mild chronic inflammatory cell infiltrate and areas of haemorrhage. Final diagnosis was made as unicystic ameloblastoma of the luminal variant (Figure 2(a) & (b)). Patient had an uneventful postoperative recovery with no signs of recurrence during the 1 year follow up examination.

#### DISCUSSION

Unicystic ameloblastoma, a variant of ameloblastoma;

requires separate consideration based on clinical, radiological and pathologic features and in its response to treatment. Whether UA originates de novo as a neoplasm or whether it is a result of neoplastic transformation of non neoplastic cyst epithelium has long been debated. Both mechanisms probably occur, but proof, which is involved in individual patient, is virtually impossible to obtain.<sup>2</sup> Clinically and radiologically UA shows characteristics of an odontogenic cyst like dentigerous, radicular, glandular odontogenic cyst, but in histologic examination shows a typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor proliferation.<sup>5</sup>

Although, most studies on periradicular lesions focus on radicular cysts and granulomas which are highly prevalent periapical lesions, nevertheless, the occurrence of non inflammatory pathoses in this area should always be kept in mind from a differential diagnosis point of view. Radiolucent lesions in the mandibular and maxillary areas surrounding the root apices might lead to a misdiagnosis of apical periodontitis, and several other pathologies such as periapical osseous dysplasia, simple bone cysts, giant cell granulomas, keratocystic odontogenic tumors, UA's, and metastatic lesions.<sup>6</sup> As these lesions present a different prognosis they should always be considered while arriving at a diagnosis. They are, however, rare. The unicystic ameloblastoma presents a special concern in this respect, being locally aggressive and nonresponsive to root canal treatment or tooth extraction.

Generally, apical inflammatory lesions appear as radiolucent images in intimate contact with the apical root. On radiographic examination, radicular cysts frequently show an oval outline with well-defined limits and uniform, symmetrical concentric growth. Additionally, alveolar cortical bone appears gradually dislocated from the insertion point of the lesion on the dental apex. Although root resorption is frequently associated with chronic periapical lesions, it is not commonly seen in small lesions and is rarely advanced.<sup>7,8</sup> Moreover, clinical manifestations of cystic lesions include slow asymptomatic growth, needle aspiration usually liberates serous fluid.<sup>8,9</sup>

Some of these clinical and radiographic features are common to UA.<sup>10</sup> Most UAs have been associated with an expansile unilocular radiolucency with root resorption but not with cortical erosion and perforation.<sup>11,12</sup> Moreover, pulp sensitivity testing is important for the differential diagnosis between apical lesions of endodontic and nonendodontic origin. In the present case, carious #21 and #22 along with the radiographic findings gave weightage to a diagnosis of

periapical cyst but the microscopic examination showed an entirely different picture, suggesting to the practitioner to keep in mind the differential diagnosis of apical lesions, and the presence of a noncystic lesion.

On comparing UA with a solid multicystic ameloblastoma, some authors believe that this lesion is less aggressive than its solid or multicystic form and even curettage has been performed as the indicated therapeutic approach. The age of the patient is another influencing factor related to the choice of treatment.<sup>13</sup> As UA tends to affect young adolescent patients, the concern to minimize surgical trauma and permit jaw function and tooth development to proceed reasonably unimpaired should be one of the important aspects in tumor management. While conservative surgery seems to have been justified in preference to mutilating radical surgery for the young patients, choice of treatment has to be considered in conjunction with other clinical and pathological factors such as the size, location and growth pattern of the tumor. Whatever surgical approach the surgeon decides to take, long term follow up is mandatory, as recurrence of unicystic ameloblastoma may be long delayed.<sup>5</sup>

The importance of UA is that it possesses a much better prognosis after enucleation or curettage than does the classic intraosseous ameloblastoma. Its recurrence rate after these procedures is around 15%, whereas recurrence is much higher after curettage of classic intraosseous ameloblastomas. The reason for this better prognosis is that in many examples the ameloblastoma involves only the epithelial lining of the cyst or projects into its lumen, and the histopathological findings of the present case is in concordance with it. These lesions, which are sometimes referred to as luminal and intraluminal ameloblastomas, respectively, are confined by the fibrous connective tissue wall of the cyst and are consequently removed completely if the cyst is enucleated.<sup>2</sup>

## CONCLUSION

Unicystic ameloblastoma can be found at or near tooth apices, simulating a radicular cyst or adenomatoid odontogenic tumor or some other non inflammatory pathosis. So diagnosis should be based on correlation of clinical, radiographic and histopathological features to clear any doubts regarding the ambiguous nature of such lesions which are found in the peri-radicular region.

## REFERENCES

1. Manchanda A, Narang R, Singh B. Odontogenic tumors: pathogenesis and historical update-A review. *Ind J Comp Dent Care* 2014; 4: 76-82.
2. Paikkatt V, Sreedharan S. Unicystic ameloblastoma of the

- maxilla: a case report. *J Indi Soci Pedo Prev Dent* 2007; 25: 106-110.
3. Philipsen HP, Reichert PA. Classification of odontogenic tumours. A historical review. *J Oral Pathol Med* 2006; 77: 85.
  4. Cunha E, Fernandes A. Unicystic ameloblastoma: a possible pitfall in periapical diagnosis. *Int Endo J* 2005; 38: 334-340.
  5. Satyabhushan N. Unicystic ameloblastoma masquerading as huge periapical lesion, both clinically and histopathologically: two case reports with review of literature. *J Clin Imag Sci* 2013; 3: 9.
  6. Gondak R. Unicystic ameloblastoma mimicking apical periodontitis: a case series. *J Endo* 2013; 39: 145-147.
  7. Wood NK, Goaz PW. Differential diagnosis of oral and maxillofacial lesions. Chapter 15, 3rd edn. St Louis, MO, USA: Mosby, pp: 320-56.
  8. Neville BW, Damm DA, Allen CM, Bouquot JE. Oral and maxillofacial pathology 2002; Chapter 15, 2<sup>nd</sup> edn. Philadelphia, USA; WB Saunders company, pp: 616-8.
  9. Shear M. Cysts of the oral regions. Chapter 11, 2<sup>nd</sup> edn. Oxford, UK: Wright, pp: 281-97.
  10. Eversole LR, Leider AS, Hansen LS. Ameloblastoma with pronounced desmoplasia. *J Oral Maxillofac Surg* 1984; 42: 735-40.
  11. Li TJ, Wu YT. Unicystic Ameloblastoma: a clinicopathologic study of 33 Chinese patients. *Am J Surg Pathol* 2000; 24: 1385-92.
  12. Li TJ, Kitano M, Arimura K, Sugihara K. Recurrence of unicystic ameloblastoma: A case report and review of the literature. *Arch Pathol Lab Med* 1998; 122: 371-74.
  13. Robinson L, Martinez MG. unicystic ameloblastoma: A prognostically distinct entity. *Cancer* 1977; 40: 2278-85.