

ORAL PATHOLOGY

UNICYSTIC AND SOLID TYPE OF AMELOBLASTOMA OCCURRING IN SAME PERSON

A RARE CASE



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Abstract

We present a case of unicystic ameloblastoma with mural proliferation in anterior mandible and solid/multicystic ameloblastoma with plexiform pattern in posterior mandible in a 29 year old male patient. The possibility may be, these two are completely separate lesions or they may be due to same lesion showing skip bone involvement.

Key words: ameloblastoma, unicystic ameloblastoma, mural

Introduction

Ameloblastoma is a benign, locally aggressive odontogenic neoplasm with variable clinical expression and accounts for 1% of all cysts / tumors of the jaws and 18% of all odontogenic neoplasms.¹ It is typically slow growing, locally aggressive and rarely metastasizes, but has a high rate of recurrence (55-90%) if not removed adequately.² As per the WHO system of 2003, ameloblastoma is included under "benign neoplasms and tumor-like lesions arising from the odontogenic apparatus showing odontogenic epithelium with mature fibrous stroma, without ectomesenchyme" and is classified based on differences in biologic behavior, treatment plan and recurrence rate as follows;³

1. Classic solid / multicystic ameloblastoma.
2. Unicystic ameloblastoma.
3. Peripheral ameloblastoma.
4. Desmoplastic ameloblastoma, including so-called hybrid lesions.

The concept of unicystic ameloblastoma was introduced in 1977 by Robinson and Martinez,⁴ who considered it to be a less aggressive variant of ameloblastoma and suggested simple enucleation as treatment. However, the term unicystic ameloblastoma was adopted in the second edition of the international histologic classification of odontogenic tumors and encompasses lesions previously referred to as cystic ameloblastoma, ameloblastoma associated with dentigerous cyst, cystogenic ameloblastoma, extensive dentigerous cyst with intracystic ameloblastic papilloma, dentigerous cyst with ameloblastomatous proliferation, ameloblastoma developing in a radicular (or globulomaxillary) cyst, luminal ameloblastomas, mural ameloblastomas and ameloblastomas arising in dentigerous cysts. It refers to those cystic lesions that show clinical, radiographic or gross features of jaw cyst, but on histologic examination shows typical ameloblastomatous epithelium, lining part of the cystic cavity with or without luminal and / or mural tumor growth.⁵

Case report

A 29 year old male patient presented with history of pain and swelling with purulent discharge in left mandibular anterior region over a period of week. He had undergone extractions but did not possess records of the treatment history. As per the patient the current problem developed after the extractions as a swelling, which had progressively increased to the present size over a year's duration.

Clinical examination revealed swelling of mandible extending from angle to parasymphysis region, which was firm and non-tender. The expanded lingual

cortical plate was seen to cross the midline from 33 to 43 which led to discomfort in tongue positioning. The swelling on the buccal vestibule extended from 38 to 42 crossing the midline.

Panoramic radiograph revealed a multilocular radiolucency (Fig.1) in left body of the mandible extending from right first premolar to edentulous area in left molar region (approximate size of 7 cm X 2.5 cm). Resorption of roots was evident in relation to 31, 32, 33, 41, 42 and 43.

Based on clinical and radiographic findings, ameloblastoma was suspected. Histological examination of incision biopsy from anterior mandible revealed a fibrous cystic wall with ameloblastomatous epithelial lining. The epithelium showed basal layer of columnar cells with hyperchromatic nuclei showing reverse polarity and basilar cytoplasmic vacuolisation. The overlying epithelial cells were loosely cohesive and resembled stellate reticulum (Fig.2). Based on these findings, case was diagnosed as unicystic ameloblastoma subgroup-1. A conservative treatment plan of enucleating the lesions was advocated and implemented.

On surgical exploration presence of intact bone separating anterior and posterior part of the lesion was evident at premolar region (Fig.3). Histological examination of entire excisional tissue from anterior mandible at multiple levels revealed again a fibrous cystic wall with ameloblastomatous epithelial lining (Fig.4). But, ameloblastoma tissue was found to extend into the fibrous wall of cyst (Fig.5) representing features of unicystic ameloblastoma with mural proliferation i.e. subgroup-1.3. Tissue from posterior part of the mandible showed features of plexiform ameloblastoma with anastomosing cords of odontogenic epithelium. The cords were bound by columnar and cuboidal cells surrounding more loosely arranged epithelial cells (Fig.6). It was decided to monitor the case closely for recurrence instead of immediate extensive second surgery. Indeed the case showed recurrence after a year and a marginal resection was planned.

Discussion

Unicystic ameloblastoma is second important clinical type of ameloblastoma and accounts for 10-15% of all intraosseous ameloblastomas.⁶ They have been reported to occur in second and third decades of life as against its solid counterpart which occurs in fourth decade of life. Unicystic ameloblastoma most commonly occurs in posterior mandible followed by parasymphysis region, anterior maxilla and posterior maxilla.⁷

Most unicystic ameloblastomas resemble dentigerous cyst clinically and radiographically, but a few are not associated with impacted teeth which are called non dentigerous variant and resemble residual cyst. The mean age of nonimpacted tooth related cystic ameloblastoma was 35 years in comparison to 16.5 years for the impacted tooth related variant.⁸

Leider et al proposed three pathologic mechanisms for evolution of unicystic ameloblastoma⁵

- a. The reduced enamel epithelium associated with a developing tooth undergoes ameloblastic transformation with subsequent cystic development.
- b. Ameloblastomas arise in dentigerous or other types of odontogenic cysts in which the neoplastic ameloblastic epithelium is preceded temporarily by a non-neoplastic stratified squamous epithelial lining.
- c. A Solid ameloblastoma undergoes cystic degeneration of ameloblastic islands with subsequent fusion of multiple micro cysts and develops into a unicystic lesion.

Unicystic ameloblastoma must be differentiated from other odontogenic cysts as former has higher rate of recurrence than latter.⁹ Histologically minimum criterion for diagnosis of unicystic ameloblastoma is demonstration of cystic sac lined by ameloblastomatous epithelium. Ackermann et al. classified unicystic ameloblastoma into 3 histological groups.⁶

Group I : Luminal Unicystic ameloblastoma (tumor confined to luminal surface of the cyst).

Group II : Intraluminal/Plexiform unicystic ameloblastoma (nodular proliferation into the lumen without infiltration of tumor cells into the connective tissue wall).

Group III: Mural unicystic ameloblastoma (invasive islands of ameloblastomatous epithelium in the connective tissue wall not involving the entire epithelium).

The above classification was modified further by Philipsen and Reichart⁵ as

Subgroup 1 – luminal unicystic ameloblastoma.

Subgroup 1.2 – luminal and intraluminal

Subgroup 1.2.3 – luminal, intraluminal and intramural

Subgroup 1.3 – luminal and intramural.

It was suggested that subgroup 1, 1.2 were nonaggressive and could be treated by enucleation, as capsule provides adequate margin of uninvolved tissue by surrounding the cyst completely. But there is a need to follow the case for 5-10 years for recurrence. Whereas subgroup 1.2.3, 1.3 should be treated more aggressively similar to solid/multicystic ameloblastoma as the limiting barrier is lost and infiltration of tumor may reach surrounding cancellous bone. The reliable treatment then would be marginal resection following initial enucleation and curettage. But, a definitive diagnosis of unicystic ameloblastoma can only be done by thorough histological examination of entire lesion at multiple levels and cannot be predicted preoperatively on clinical or radiographic grounds. As preoperative incision biopsy is not representative of entire lesion it may result in an incorrect classification. Thus, true nature of the lesion becomes evident only after enucleation when entire specimen is available for microscopy which was true in our case.

Robinson and Martinez,⁴ Gardner and Corio¹⁰ indicated that the recurrence rate after conservative treatment for unicystic ameloblastoma was lower than those for multicystic or solid lesions. Average interval of recurrence was found to be 7 years and was found to be related to histologic subtype. Those invading fibrous wall had recurrence of 35.7%, others only 6.7%¹¹. Recurrence rates were 3.6% for resection, 30.5% for enucleation alone, 16% for enucleation followed by Carnoy's solution application and 18% by marsupialisation followed by enucleation or resection¹². Only three of seventeen cases in Robinson and Martinez's series showed recurrence after enucleation.² Shteyer et al¹³ reviewing the literature stated the recurrence rate after enucleation of what they call mural ameloblastoma was less than 10%. Reason being in most cases, the tumor is well localized by fibrous capsule. Once tumor growth crosses the capsule, it can infiltrate the surrounding cancellous bone to behave like solid/multicystic ameloblastoma.

As presence of odontogenic epithelial islands in cyst wall influences treatment outcome, it is necessary for pathologist to carefully examine cystic ameloblastoma surgical specimen for their presence. Multiple serial sections are required to be examined. So, true nature of such lesions is evident only when entire surgical specimen is available for microscopic examination.

In our case the unicystic ameloblastoma subgroup 1 was diagnosed on incision biopsy. So, enucleation of the lesion was done. The lesion on surgical exploration revealed presence of bone separating the anterior and posterior lesions. The histological examination of the enucleated tissue revealed

unicystic ameloblastoma subgroup 1.3 in anterior and solid/multicystic ameloblastoma in the posterior mandible.

Conclusion

Cases of all clinical types of ameloblastoma have been reported in literature, but rarely have we come across a case with presentation of both solid and unicystic type in same person. This case shows unicystic ameloblastoma in anterior and solid ameloblastoma in the posterior body of the mandible. The possibilities may be solid proliferation of ameloblastoma in posterior mandible might have been from mural proliferation of odontogenic tumor epithelial cells of unicystic ameloblastoma in anterior mandible. But, surgical exploration of lesion showed intact bony septa separating two areas. As the bony septa did not show the perforation, it could be considered that the solid mass may not be mural proliferation of unicystic ameloblastoma. Other possibilities may be that two lesions are completely separate lesions or they may be due to same lesion showing skip bone involvement, or both might have been solid ameloblastomas with anterior lesion undergoing cystic degeneration of ameloblastic islands with subsequent fusion of multiple microcysts developing into a unicystic lesion. The case also proves the need to study entire lesional tissue through sectioning at many levels to confirm final diagnosis of unicystic ameloblastoma, especially subgroup as it influences the treatment plan and recurrence rate.

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Figure legends

Figure.1 - Panoramic radiograph showing multilocular radiolucent lesion in left body of mandible extending from right first premolar to edentulous area in left molar region. Root resorption is evident in relation to 31, 32, 33, 41, 42 and 43.

Figure.2 - Photomicrograph showing ameloblastomatous epithelial lining- incision biopsy.

Figure.3 - Intact bone at premolar region seen at surgery separating anterior and posterior lesions (arrow).

Figure.4 - Photomicrograph showing fibrous cystic wall with ameloblastomatous epithelial lining- excision biopsy.

Figure.5 - Photomicrograph showing ameloblastoma tissue extended into the fibrous wall of cyst.

Figure.6 - Photomicrograph showing plexiform ameloblastoma at higher magnification showing cords bound by columnar and cuboidal cells surrounding more loosely arranged epithelial cells.

Figure No 1



Figure No 2

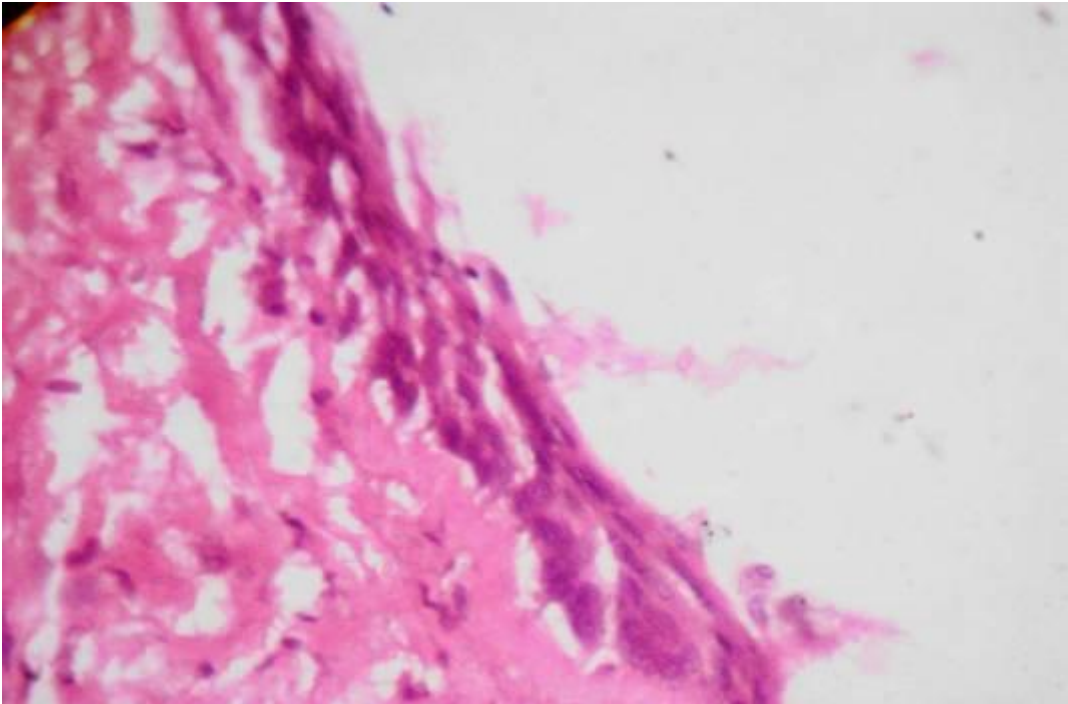


Figure No 3



Figure No 4

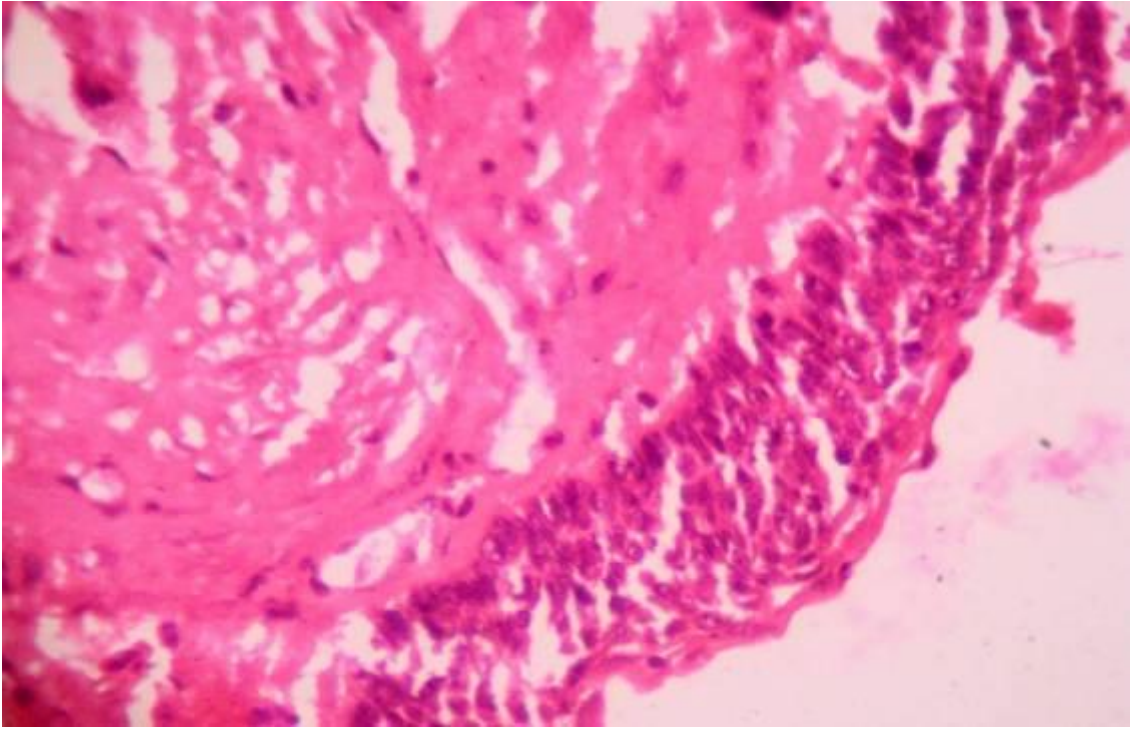


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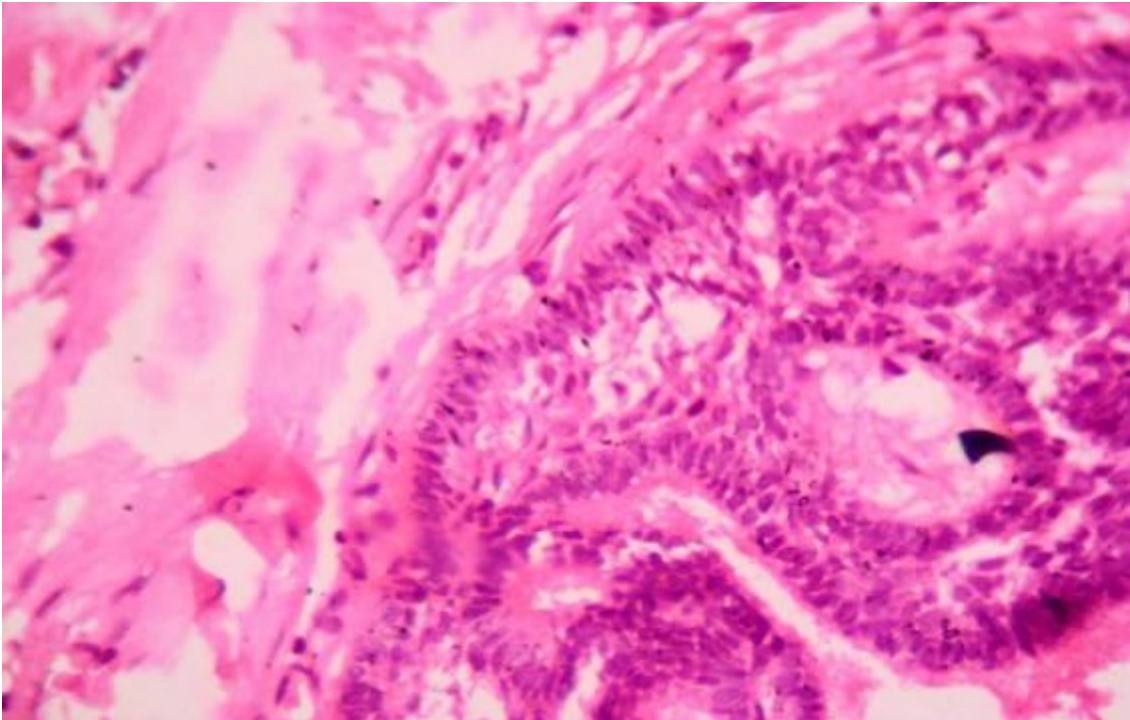


Figure No 6

