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A RARE CASE OF MULTILOCULAR UNICYSTIC AMELOBLASTOMA: A PROGRESSIVE RADIOGRAPHIC PRESENTATION.

[1]Dr. Padmashree S., [2]*Dr. Sayyad Zohabhasan, [3]Dr. Shilpa P. S., MDS and [4]Dr. Rema J.

[1]Professor and Head, Department Oral Medicine and Radiology Vydehi Institute of Dental Sciences, Bangalore, Karnataka, India.
[2]3rd Year PG Student Department Oral Medicine and Radiology Vydehi Institute of Dental Sciences, Bangalore, Karnataka, India.
[3]Senior Lecturer Department of Oral Medicine and Radiology Vydehi Institute of Dental Sciences, Bangalore, Karnataka, India.
[4]Professor, Department of Oral Medicine and Radiology Vydehi Institute of Dental Sciences, Bangalore, Karnataka, India.

Corresponding Author: Dr. Sayyad Zohabhasan
3rd Year PG student Department Oral Medicine and Radiology Vydehi Institute of Dental Sciences, Bangalore, Karnataka, India.

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ABSTRACT
Unicystic ameloblastoma (UA) refers to those cystic lesions that show clinical, radiographic, or gross features of a mandibular cyst, but with ameloblastomatous epithelium lining part of the cyst cavity histopathologically, with or without luminal and/or mural tumour growth. Among different types of ameloblastoma, unicystic ameloblastoma is a less encountered variant and radiographically appears as unilocular or multilocular radiolucency. In this article, we report a rare case of multilocular unicystic ameloblastoma, with series of radiographs showing its progression and emphasis on histopathology and management.

KEYWORDS: Unicystic ameloblastoma, unilocular, multilocular, Odontogenic tumor.

INTRODUCTION
Ameloblastoma is a most common Odontogenic tumor, arising from epithelial cellular elements and dental tissues in various phases of development.[1] It is a slow-growing, persistent, and locally aggressive neoplasm with its peak incidence in the 3rd to 4th decades of life and has an equal sex distribution.[2] The vast majority of ameloblastomas arise in the mandible, especially in the angle and ramus region. Based on clinicoradiological findings, ameloblastoma can be grouped into three forms: multicystic or solid, unicystic, and peripheral forms.[3]

Unicystic ameloblastoma (UA) is a variant ameloblastoma. The occurrence of UA has been reported to be 5 to 22% of all the types of ameloblastomas.[4] In this article, we report a rare case of multilocular unicystic ameloblastoma, with series of radiographs showing its progression and emphasis on histopathology and management.].

CASE REPORT
A 41 year old male patient reported to the Department of Oral Medicine and Radiology with a chief complaint of swelling on the right side of the face since 11 years (since the year 2004). Patient visited a local practitioner 11 years back for the same swelling, who extracted the molars in that region. After extraction patient did not undergo the surgical removal of the lesion. This was followed by progressive increase in the size of the lesion. Later, drainage and curettage was attempted after eight years (in the year 2012), following which slight reduction is the swelling was reported by the patient. Again, curettage was attempted after one year (in the year 2013) by the local practitioner, with no significant change in the swelling. After two years (in the year 2015), patient reported to our department, with the complaint of progressive increase in the size of the swelling. Patient was diagnosed with hypertension and diabetes, one month ago, but was not prescribed with any medication.

On extraoral examination there was a diffuse swelling present on the right side of the face involving middle third and lower third of the face, measuring around approximately 8 cm × 9 cm in its greatest dimension, with normal overlying and surrounding skin surface (Figure 1). On palpation swelling was firm in consistency and tender. Intra oral examination revealed mild obliteration of lower right buccal vestibule and missing mandibular right posterior teeth (teeth number 46, 47 and 48) (Figure 2). Based on the history and

clinical examination, provisional diagnosis of benign odontogenic tumour was given. Differential diagnosis was considered as Ameloblastoma, Dentigerous cyst, and odontogenic keratocyst.

Radiograph

Figure 3 (a and b) and Figure 4 (a and b) shows series of radiographic images showing the progression of the lesion over the years.

Panoramic radiograph taken eleven years (in the year 2004) back showed multilocular radiolucency with sclerotic and scalloped outline extending from mesial aspect of 47, posteriorly 1.5 centimetre anterior to the posterior border of the ramus, superiorly 1.5 centimetre below the sigmoid notch, inferiorly 1.5 centimetre above the lower border of mandible. There is root resorption of mesial root of 46. Honeycomb loculations were evident in the periapical region of 47 (Figure 3a).

After eight years (in the year 2012), panoramic radiograph shows missing 46, 47 and 48; the ostelytic lesion has extended anteriorly to the distal aspect of 45 and superiorly to involve sigmoid notch and coronoid process. A combination of honey comb and soap bubble pattern of multilocularity can be appreciated in the missing region of 46, and 47 while scalloped margins was appreciated in the ramus region. (Figure 3b).

After one year (in the year 2013), lesion has extended to involve posterior margin of ramus, with intact cortical border, destroying the coronoid process, inferiorly sparing 0.5 cm of inferior border of mandible. The internal aspect shows soap bubble pattern with minimal honey comb pattern in the missing 46, 47 region (Figure 4a).

After two years (in the year 2015), osteolytic lesion has completely destroyed posterior border of ramus with thinning of inferior border of mandible. Internal aspect shows thin remnant septae giving soap bubble appearance. (Figure 4b).

Based on these features, radiographic diagnosis of ameloblastoma was considered. The case was treated by unilateral mandibular resection. Histopathological examination showed non keratinized odontogenic epithelium with columnar and cuboidal basal cell layer showing hyperchromatic palisading nuclei with reversal of polarity. The epithelium shows extension into the underlying connective tissue with tall columnar cells with reversal of polarity seen in the connective tissue stroma. All the features were in the favour of luminal and intra mural unicystic ameloblastoma. The mandible was reconstructed using the fibular free flap under general anesthesia.

Figure legends

Figure 1: Diffuse swelling involving right side of the face

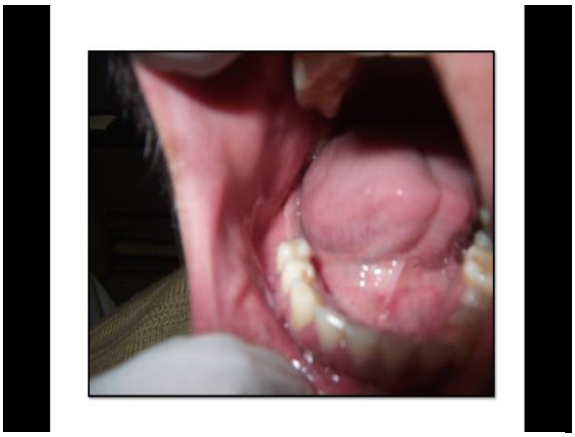

Figure 2: Intra oral examination showing mild obliteration of lower right buccal vestibule and missing mandibular right posterior teeth

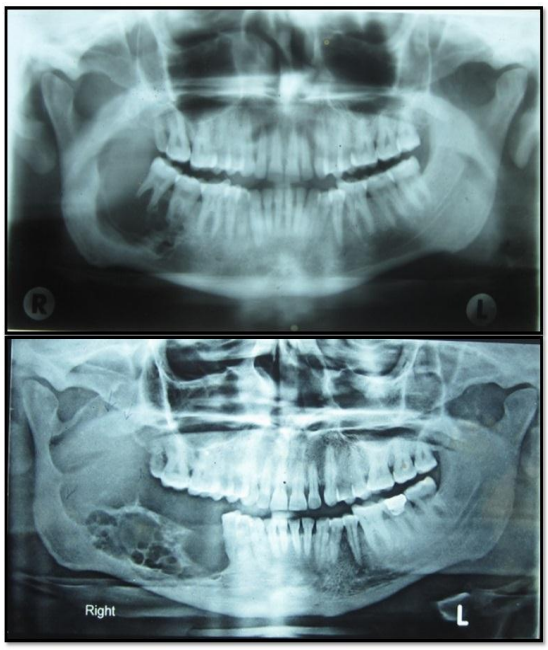

Figure 3a: Panoramic radiograph in the year 2004
Figure 3b: Panoramic radiograph in the year 2012

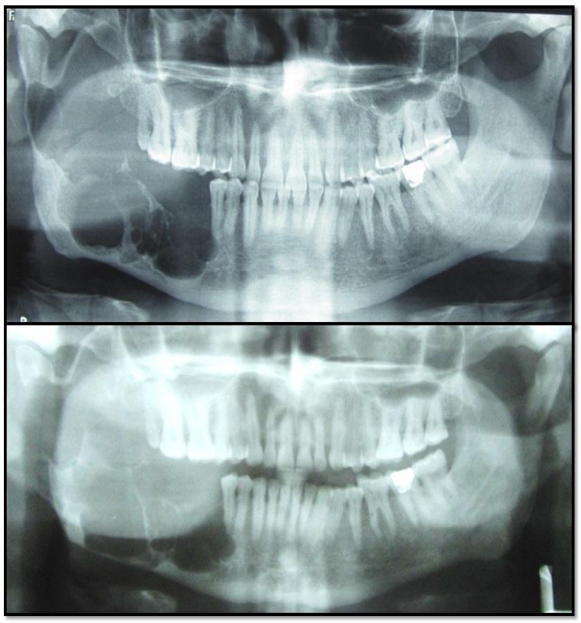

Figure 4a: Panoramic radiograph in the year 2013
Figure 4b: Panoramic radiograph in the year 2015

DISCUSSION

UA is a variant of ameloblastoma, developing within the lining, lumen, or wall of a cyst and show clinical, radiographic, or gross features of a mandibular cyst, but with ameloblastomatous epithelium lining part of the cyst cavity histopathologically, with or without luminal and/or mural tumour growth. The UA was recognised as a distinct entity by Robinson and Martinez in 1977. 46% of the cases of UA have been reported to occur in second decade of life (average – 25.5 years) with male predominance.[5,6] However, our patient was 41 year old male patient, with age of occurrence more than the general agreement. Most common site of occurrence is in posterior mandible followed by parasymphysis region, anterior maxilla and posterior maxilla.[3] Three mechanisms have been proposed for the development of unicystic ameloblastoma by Leider et al,[7] they are,

1. From reduced enamel epithelium.
2. From dentigerous cyst.
3. Cystic degeneration of solid ameloblastoma.

Radiographically unicystic ameloblastoma appears as well circumscribed unilocular radiolucency, but sometimes may also rarely have multilocular pattern. Eversole et all described six radiographic features of unicystic ameloblastoma: Unilocular, macromultilocular, pericoronal, interradicular, scalloped or periapical expansile radiolucencies.[8] Our case showed multilocular pattern 11 years back, later extending to anterior region with honeycomb pattern of multilocularity. At the time of treatment, the lesion had extended to destroy entire ramus and lower border of mandible and soap bubble pattern with thin septe in the body of the mandible.

Our case is of particular interest that the series of radiographs taken over span of 11 years, showed the progression of lesion from honeycomb pattern to combination of honeycomb and soap bubble pattern and finally to a predominantly to a soap bubble appearance. Later these locules were destroyed or coalesced leaving cystic spaces with thin remnant septae.

Ackermann classified unicystic ameloblastoma into three histologic variants: luminal with tumor confined to luminal surface of cyst (Group I); intraluminal where there is nodular proliferation into the lumen without connective tissue wall involvement (Group II) and; Mural where ameloblastomatous epithelium invade connective tissue wall not involving the entire epithelium (Group III).[9]

Philipsen and Reichart subgrouped into following types:[10]
Subgroup 1: Luminal UA
Subgroup 1.2: Luminal and intraluminal
Subgroup 1.2.3: Luminal, intraluminal, and intramural
Subgroup 1.3: Luminal and intramural

Based on these histological types, our patient showed subgroup 1.3 which required aggressive treatment. The treatment is based on clinical behaviour and histological subtype. For the luminal type where there is no extension into adjacent tissue, enucleation generally suffices. If there extension of tumor into the wall and adjacent connective tissue, as in mural type, bony resection may be necessary.

In our case, lesion progressed over 11 years, starting as multilocular lesion in the ramus, with continued destruction of mandible at the time of treatment. This shows timely intervention can prevent the lesion spread and preventing destruction of normal tissues and reduced postoperative morbidity. The patient should be followed at regular intervals for the recurrence. Although unicystic ameloblastomas are considered to be less aggressive form as compared to solid variety, recurrences are more common in those histologic subtypes with invading fibrous wall (35.7%).[11]

CONCLUSION

It can be concluded that at present, histologic examination is the most sensitive tool for differentiating between odontogenic cysts and UAs. However, both clinical and radiologic findings share equal contribution to the final diagnosis. In our present case, the progression of the lesion and radiographic features over the years has been highlighted. Thus, correlation of histopathologic findings and radiological findings are very important to arrive at definitive diagnosis. Final diagnosis may alter the therapeutic decision significantly, as all these lesions have different prognosis.

REFERENCES

1. Handa H, Bailoor DN, Naidu G, Shrivastava K, Raghuvanshi. Unicystic Ameloblastoma of mandible. Aggresive treatment A myth or a need. Case report and Extensive review of literature. IOSR Journal of Dental and Medical Sciences (IOSR-JDMS) e-ISSN: 2279-0853, p-ISSN: 2279-0861, Nov- Dec. 2013; 12(2): 26-31.
2. Gerzenshtein J, Zhang F, Caplan J, Anand V, Lineaweaver W: Immediate mandibular reconstruction with microsurgical fibula flap transfer following wide resection for ameloblastoma. J Craniofac Surg, 2006; 17(1): 178-82.
3. Philipsen HP, Reichart PA. Classification of odontogenic tumors and allied lesions. Odontogenic tumors and allied lesions Quintessence Pub. Co. Ltd 2004, 21-3 Quintessence Publication. Co. Ltd London, 2004; 77-86.
4. Robert EM, Diane S. Oral and Maxillofacial Pathology- A Rationale for Diagnosis and Treatment. Chicago: Quintessence Pub, 2003; 637-703.
5. Babu N, Charles NSC, Runwal SH. Unicystic Ameloblastoma of Mandible Treated with an Innovative Approach: A Clinical Case Report J Clin Diagn Res, Jul, 2015; 9(7): ZD11–ZD13.
6. Gupta N, Saxena S, Rathod VC, Aggarwal P. Unicystic ameloblastoma of the mandible. J Oral Maxillofac Pathol, May-Aug, 2011; 15(2): 228–231.
7. Leider AS, Eversole LR, Barkin ME. Cystic ameloblstoma. Oral Surg Oral Med Oral Pathol, 1985; 60: 624–30.
8. Eversole, LR, Leider AS, Strub D. Radiographic characteristics of cystogenic ameloblastoma. Oral Surg Oral Med. Oral Pathol, 1984; 57(5): 572-7.
9. Ackermann GL, Altini M, Shear M. The unicystic ameloblastoma: A clinicopathological study of 57 cases. J Oral Pathol, 1988; 17: 541–6.
10. Reichart, P. A. & Philipsen, H. P. Odontogenic tumors andallied lesions. Hanover, Quintessence Publishing Inc, 2004; 69-86.
11. Li T, Wu Y, Yu S, Yu G. Clinicopathological features of unicystic ameloblastoma with special reference to its recurrence. Zhonghua Kou Qiang Yi Xue Za Zhi, 2002; 37: 210–2.