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Is There a Correlation Between Radiographic Presentation of Multilocularity and Aggressive Histology of Unicystic Ameloblastomas. – Report of Two Cases and Review of Literature

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Authors' contributions

This work was carried out in collaboration among all authors. Author SS designed the study and performed the statistical analysis. Author PG wrote the protocol and wrote the first draft of the manuscript and author SN managed the analyses of the study. Authors SP, NS and PB managed the literature searches. All authors read and approved the final manuscript.

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Case Study

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ABSTRACT

Conventional or Solid/ Multicystic Ameloblastomas (SMA) are conventionally treated with resection while Unicystic Ameloblastoma (UA) is usually treated conservatively. Mural variety of UA has shown to be more aggressive and behaves similar to conventional ameloblastoma and is therefore advised to be treated aggressively. So treating a mural variety of unicystic ameloblastoma conservatively can lead to recurrence. Recurrence of multilocular ameloblastoma is high as compared to unilocular if treated conservatively. Does this hold true for multilocular unicystic ameloblastoma. Also, do multilocular ameloblastoma show aggressive mural type of lining on histopathology. We present two cases of multinodular unicystic ameloblastoma and try to correlate if multilocular unicystic ameloblastoma show aggressive histology from literature review.

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Keywords: Unicystic ameloblastoma; multilocular radiolucency; mandible; maxilla; mural.

ABBREVIATIONS

UA : Unicystic Ameloblastoma,
SMA : Solid/Multicystic Ameloblastomas

1. INTRODUCTION

The UA is recognized as a distinct subtype of Ameloblastoma based on clinical and radiological features as well as distinct histopathological findings [1]. It is mostly unilocular radiologically and lining shows features of ameloblastoma in luminal, intraluminal and mural pattern. On the contrary, Conventional or SMA usually show a mutlilocular/ soap bubble appearance on radiographs with follicular, plexiform, acanthomatous, granular, basaloid and desmoplastic features on histology. SMA's are conventionally treated by resection while UA is usually treated conservatively. Mural variety of UA behaves aggressively similar to SMA and is accountable to be treated aggressively [2]. Incisional biopsy is not a true representative of the lesion in case of UA due to varied presentation of its lining including invasion of connective tissue. So treating a mural variety of UA conservatively can lead to recurrence. It is suggested to examine the specimen after enucleation to determine the need for further treatment which involves second surgery [2]. A simpler way is needed to determine which UA needs aggressive therapy and which can be treated conservatively. Recurrence of multilocular ameloblastoma is high as compared to unilocular if treated conservatively [3]. Does this hold true for multilocular unicystic ameloblastoma? Also, do multilocular unicystic ameloblastoma show aggressive mural type of lining on histopathology? We present two cases of multilocular UA and review the literature to find the answers.

2. CASE REPORTS

2.1 Case Report 1

A 25-year-old male patient reported to our department with a chief complaint of a gradually increasing swelling over the anterior lower jaw since 4 months. On examination, diffuse swelling was seen extending from right para-symphysis to left angle of mandible (Fig. 1). Appreciable expansion of the buccal and lingual cortices was present. Intraorally swelling was firm on palpation along the obliterated vestibule and revealed multiple small areas of soft compressible, fluctuant mucosal thinning suggestive of cortical perforations. Grade 1 Mobility was noted with mandibular anterior teeth and the bilateral 1st and 2nd premolars.

Fig. 1. Pre-operative (extra-oral view) image showing extension of the swelling and asymmetry

OPG revealed a large multi-locular radiolucency extending from left mandibular angle region crossing the midline to extend up to the right mandibular body region. Root Resorption of the associated teeth was also present. 3D-Computed tomography revealed lytic expansile lesion showing bucco-lingual cortical expansion and perforation at multiple sites along with presence of multiple septae (Fig. 2). Inferior border expansion and perforation was also evident.

An Incisional biopsy was undertaken. Soft tissue sections revealed non keratinized cystic lining of uniform thickness with basal columnar cells resembling Ameloblastoma. Also, intraluminal proliferation of Ameloblastomatous epithelium in a plexiform pattern was seen. At certain regions the cystic lining was breached showing downward proliferation of basal cells and formation of Ameloblastic follicles. These features were consistent with a diagnosis of mixed variant of unicystic ameloblastoma. Due to young age of patient and extensive bony involvement, Marsupialization of the lesion was

attempted, but the lesion did not show signs of regression even after few months. Subsequently resection of the mandible from right body to the left angle region and reconstruction with Microvascular Free Fibula Flap was undertaken (Fig. 3). Final histopathology of the resected specimen confirmed the diagnosis of mixed variant UA (Fig. 4). Patient is free of recurrence at 15 months follow up.

2.1 Case Report 2

A 42-year-old female reported to the outpatient department with chief complaint of gradually increasing swelling in the upper anterior maxilla of 6 months duration with mobility and exfoliation of upper left three anterior teeth (Fig. 5). Also the patient had history of trauma to the same region 12 years back.

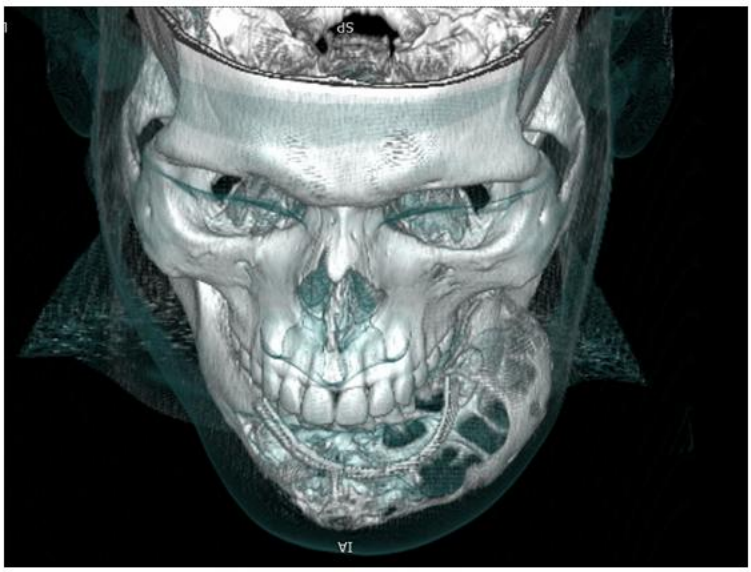

Fig. 2. 3D-Computed tomography showing bucco-lingual cortical expansion with inferior border involvement and perforation at multiple sites along with presence of multilocular appearance due to multiple septae in the mandible

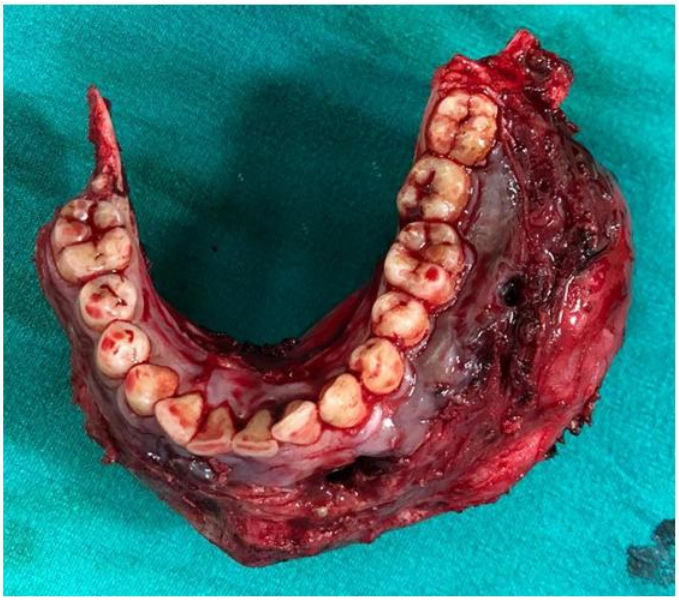

Fig. 3A and B. Resection of the mandible followed by reconstruction by microvascular free fibula flap

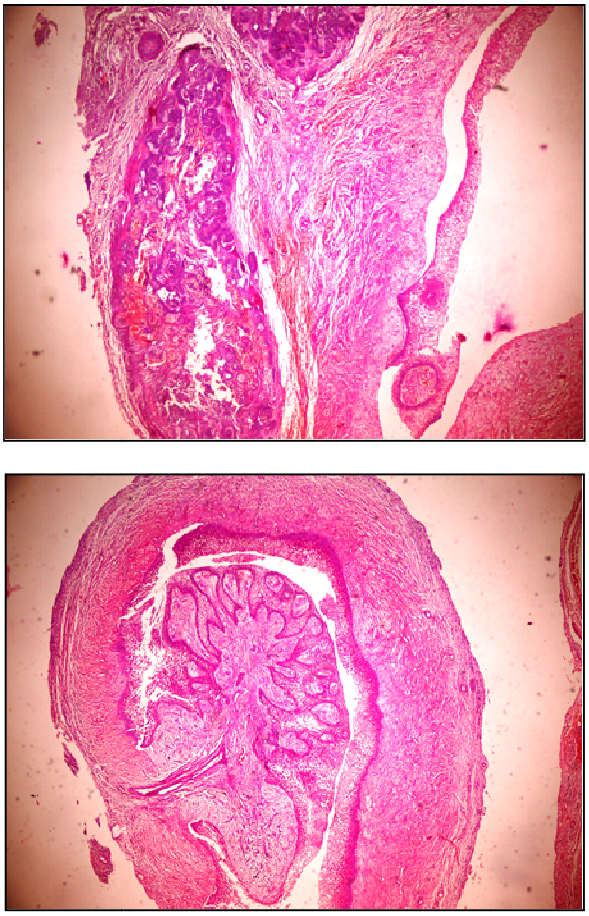

Fig. 4. H&E Stain (10 X Magnification) a) shows the luminal and intraluminal proliferation and b) mural growth of ameloblastic follicle

Fig. 5. Pre-operative (extra-oral view) image showing extension of the swelling and asymmetry

On intraoral examination, diffuse swelling was seen buccally and palatally extending from right incisor region to the left molars. Few missing anteriors were noted. On aspiration, 1-1.5ml of red tinge colored fluid was obtained conferring to a diagnosis of Cyst. Orthopantomogram and Cone Beam Computed Tomography revealed a mixed, multilocular lesion crossing the midline extending from 11 to 25, encroaching the middle meatus superiorly and alveolar crest inferiorly (Fig. 6). Incisional biopsy was suggestive of Acanthomatous Ameloblastoma. A Left Extended Infrastructure Hemi-Maxillectomy was performed using the midfacial degloving approach (Fig. 7). Final histopathology of the resected specimen was a mixed variant of UA (Fig. 8). Post operatively at 15 months follow up patient is free of recurrence.

3. DISCUSSION

UA is a rare variant of Ameloblastoma, believed to have good prognosis when compared with traditional solid variety. UA tends to affect younger age groups in contrast to conventional type, with about half of cases occurring in the second decade of life, few cases do occur in a much older age group[3] equivalent to our experience. UA is most commonly seen in posterior mandible in association with unerupted third molar. In maxilla, it is mostly seen in posterior region [2].

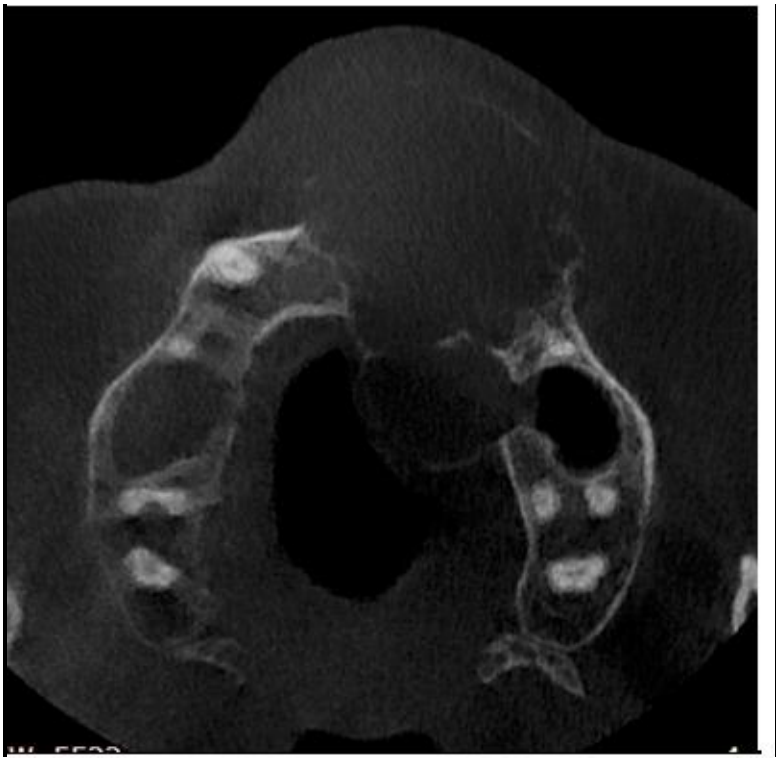

Fig. 6a.

Fig. 6b.

Fig. 6A and B: Axial and Coronal sections of Cone Beam Computed Tomography revealed a multilocular lesion crossing the midline extending from 11 to 25, encroaching the meatus superiorly and alveolar crest inferiorly

UA has been divided into three categories according to the pattern of proliferation of Ameloblastomatous epithelium as Luminal (type 1), Intra-luminal (type 2) and Mural (type 3) [4,5]. A Conventional Ameloblastoma is usually a multilocular radiolucent lesion and unilocular radiolucency is less common.

A diagnosis of UA is made if there is a well-defined radiolucent lesion which is mostly unilocular. According to literature, variants of unicystic ameloblastoma can show six radiographic patterns including multilocular appearance [6]. Also there is 'dentigerous' and 'non dentigerous' variety of UA depending on presence or absence of impacted tooth [5].

Radiographically, unilocular to multilocular pattern is seen in a ratio of 8:7 for cases of UA without impacted teeth when compared to 13:3 for UA associated with impacted teeth [5] Both Eversole and Leider [6,7] found that Unicystic Ameloblastomas when associated with a retained tooth showed a much lower recurrence rate.

Lau et al. conducted a systematic review on recurrence related to treatment modalities of Unicystic Ameloblastoma [8]. They concluded that there was weak evidence that excision lead to low recurrence but advised treatment to be individualized. In review by Philipsen [5], it is clearly stated that radiological appearance of ameloblastoma may further influence the

therapeutical approaches. Conservative therapy of multilocular ameloblastomas revealed a recurrence rate of 30% in contrast to 19.2% for radical therapy [3]. Also multilocular variant results in 24% recurrence as compared to 16% recurrence in case of unilocular variant [5]. But does the multilocular unicystic ameloblastoma show aggressive histology of mural variant as compared to unilocular UA in the non impaction as compared to with impacted teeth. Can multilocular lesion showing luminal variety of UA on biopsy be treated aggressively in the first stage as SMA instead of planning a second operation after enucleation. We reviewed the literature to find out if any author has studied the radiological and histological aspects of UA and correlated the findings. (Table 1).

We could find only 4 articles where radiographic pattern of unilocular and multilocular appearance were recorded for Unicystic Ameloblastoma. Only 2 articles recorded the histology in relation to radiographic pattern.

In Rosenstein et al. [9] article on cystic ameloblastoma 6 out of 21 lesions were multilocular and 50% showed mural proliferation. In their study 9 out of 21 showed recurrence and 5 out of these 9 had mural proliferation. So 4 did not show any mural or luminal proliferation. Were these unilocular or multilocular is not detailed in the article. The authors are of the view that cystic ameloblastomas are frequently multilocular and may exhibit clinical aggressiveness.

Lawal et al. [10] in review of 15 Unicystic Ameloblastoma, radiographs were available for 12 cases. In this 8:4 ratio of multilocular to unilocular variant were noted. Of the 8 multilocular, mural variety was seen in 2, luminal variety in 2 and 4 had intraluminal variant of UA.

Lie Jun li et al. [11] reviewed 33 Unicystic Ameloblastoma. out of 33, 7 were multilocular. 35% of 17 cases out of 29 which were followed up for more than 4 years recurred. Again no correlation between locularity and recurrence assessed.

Fig. 7. Extended Hemi-maxillectomy specimen

Table 1. List of cases

Author	No. of cases	Location	Multilocular	Histology in relation to multilocularity	Histology in general	Recurrence in general	Associated impacted teeth with multilocularity
Rosenstein et al	21	Mandible -21	6	Mural or luminal -3 Cystic -3	Mural -10 Cystic -7 Intraluminal-4 9 cases recurred.	9	No data
Lawal et al	15	Mandible- 14 Maxilla -1	8	Intraluminal-4 Luminal-2 Mural -2	Mural -6 Luminal-4 Intralumnal-5	No recurrence as aggressive treatment undertaken	1
Tie Jun li et al	33	Mandible-30 Maxilla-3	7	No data available	Luminal-8 Intraluminal-10 Mural -15	6 cases recurred after 4 years. No recurrence at 4 years	19 cases out of 33 had impacted teeth but no correlation with multilocularity
Leider et al	31	Mandible -31	10	No data	Mixed pattern in 53%	Follow up period 2-5 yrs 3 out of 14 recurred	16

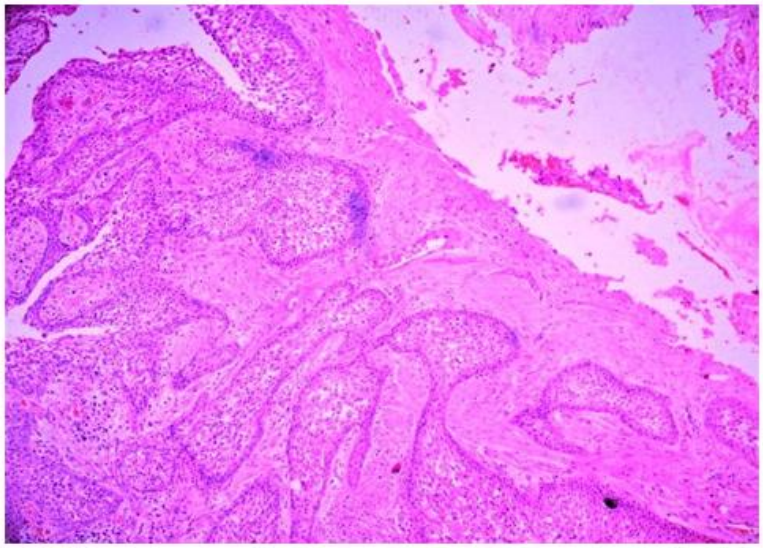

Fig. 8. Intramural proliferation of ameloblastomatic follicles evident. The thin connective tissue capsule shows collagen fibres with absence of chronic inflammatory cells. (H&E 100x).

In Clinicopathologic analysis of 31 cases of Unicystic Ameblastoma by Leider et al. [7], 10 were multilocular radiographically, mixed pattern of histology was seen in 53 % of 31 cases but no differentiation was made whether they were seen in unilocular or multilocular variant. 11 cases were followed for less than 2 years and there was no recurrence noted. 3 of the 14 cases followed for 2-5 years recurred. None of these cases were associated with impacted teeth.

In our cases, one lesion was in mandible and second was in maxilla. Both the lesions were multilocular. First case showed mixed variant UA on incisional biopsy and the second one was suggestive of Acanthomatous Ameloblastoma. Both the lesions were treated radically and final histology was consistent with mixed variety of UA.

4. CONCLUSION

There is not much clarity from the literature on the recurrence rate of unilocular vs multilocular UA as this aspect has not been studied. But limited literature evidence and reviews are suggestive of aggressive treatment for cases of multilocular variant of unicystic ameloblastoma especially with a non impacted variety. Future research is needed to study and correlate the radiographic and histologic aspect of Unicystic Ameloblastoma and the incidence of recurrence in relation to conservative and radical therapy to guide proper treatment.

CONSENT

As per international standard or university standard, patient's written consent has been collected and Images taken with the patient's consent which has been preserved by the author(s).

ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

COMPETING INTERESTS

Authors have declared that no competing interests exist.

REFERENCES

1. Robinson L MM. Unicystig ameloblastoma. A prognostically distinct entity. Cancer. 1977;40:2278–85.
2. El-Naggar AK, Chan JKC, Grandis JR, et al. WHO Classification of Head and Neck Tumours. 4th Edition. IARC: Lyon. 2017;347.
3. Reichart PA, Philipsen HP, Sonner S. Ameloblastoma: Biological profile of 3677 cases. Eur J Cancer Part B Oral Oncol. 1995;31:86–99.

4. Ackermann GL, Altini M, Shear M. The unicystic ameloblastoma: a clinicopathological study of 57 cases. J Oral Pathol Med. 1988;17:541–6.
5. Philipsen HP, Reichart PA. Unicystic ameloblastoma. A review of 193 cases from the literature. Oral Oncol. 1998;34:317–25.
6. Eversole LR, Leider AS, Strub D. Radiographic characteristics of cystogenic ameloblastoma. Oral Surgery, Oral Med Oral Pathol. 1984;57:572–7.
7. Leider AS, Eversole LR, Barkin ME. Cystic ameloblastoma. A clinicopathologic analysis. Oral Surgery, Oral Med Oral Pathol. 1985;60:624–30.
8. Lau SL, Samman N. Recurrence related to treatment modalities of unicystic ameloblastoma: a systematic review. Int J Oral Maxillofac Surg. 2006;35:681–90.
9. Rosenstein T, Pogrel MA, Smith RA, Regezi JA. Cystic ameloblastoma—Behavior and treatment of 21 cases. J Oral Maxillofac Surg. 2001;59:1311– 1316.
10. Lawal AO, Adisa AO, Olajide MA. Cystic ameloblastoma: a clinico-pathologic review. Annals of Ibadan Postgraduate Medicine. 2014 Sep 15;12(1):49-53.
11. Li T-J, Wu Y-T, Yu S-F, et al. Unicystic ameloblastoma. A clinicopathologic study of 33 Chinese patients. Am J Surg Pathol. 2000;10:1385–1392.

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