Case Report

JRAD Journal of Research and Advancement in Dentistry

J Res Adv Dent 2017;6:3:193-197.

Unicystic Ameloblastoma: A Brief Review of Literature

Rajdeep Das[1*] Rajarshi Bandyopadhyay[2] Sudipta Sahu[3] Abhinaba Bose[4] Mayukh Misra[5]

[1]Senior Lecturer, Department of Oral Pathology, Haldia Institute of Dental Sciences and Research, Haldia, West Bengal, India.
[2]Professor and Head, Department of Oral and Maxillofacial Surgery, Haldia Institute of Dental Sciences and Research, Haldia, West Bengal, India.
[3]Senior Lecturer, Department of Oral and Maxillofacial Surgery, Haldia Institute of Dental Sciences and Research, Haldia, West Bengal, India.
[4]Senior Lecturer, Department of Oral and Maxillofacial Surgery, Haldia Institute of Dental Sciences and Research, Haldia, West Bengal, India.
[4]Reader, Department of Oral and Maxillofacial Surgery, Haldia Institute of Dental Sciences and Research, Haldia, West Bengal, India.

ABSTRACT

Background: Ameloblastoma is the most common odontogenic tumour, infamous for recurrence and invasive growth. Among its various types multicystic ameloblastoma is the most frequently encountered subtype,whereas unicystic ameloblastoma is a variant of solid or multicystic ameloblastoma. The clinical and radiologic features can mimic cystic lesions of odontogenic or inflammatory origin. Keeping this point in mind it becomes essential for us to make correct diagnosis of the lesion with the help of histopathologic evaluation, which will lead to proper planning of treatment modalities thereby lowering chances of recurrence of the lesion.

Keywords: Mandible, Unicystic ameloblastoma, plexiform ameloblastoma, follicular ameloblastoma, odontogenic tumour.

INTRODUCTION

Ameloblastoma is the most commonly encountered tumour of odontogenic origin[1]. The neoplasm was first described by Cusak in 1827[2]. The name comes from old french word "Amel" meaning enamel and greek word "Blastos" meaning germ or bud[3]. The term ameloblastoma was coined by Ivey and Churchill in 1930, a currently accepted term[4][5]. According to the definition given by World Health Organisation it is locally invasive polymorphic neoplasia that often has a follicular or plexiform pattern in a fibrous stroma[6].

CASE REPORT

A 60 years old female patient came to our department with complaint of swelling in right back region of lower jaw since 06 months. The family history and personal history did not revealed any significant information. She underwent surgery for the same twice in separate institutions. The patient was of average build, well nourished with normal gait and vital signs. A diffuse swelling was seen in lower right one third of face measuring approximately 1cmx 1cm diameter extending from 1cm away from angle of mouth to 2cm before angle of mandible, mesiodistally and 1cm above the lower border of mandible, superoinferiorly(fig.1). Intra oral examination revealed buccally tilted 46 and lingually tilted 47 along with buccal and lingual cortical plate expansion(fig.2). Radiographs revealed unilocular radiolucency with buccolingual cortical plate expansion (fig.3, 4). On aspiration straw cloured fluid was obtained but cytology was non conclusive. Dentigerous cyst was considered for provisional diagnosis. The cystic specimen obtained was of dimensions 4x2.5x2 cm(fig.5). The histopathologic examination demonstrated the presence of lining epithelium of varying thickness(fig.6). At many fields cells were showing tall columnar appearance with polarized nucleus and superficial cells resembled the stellate reticulum like cells(fig.7). Invading islands of ameloblastoma were identified within the connective tissue capsule. Plexiform pattern of proliferation of the lining epithelium was identified

Received: Feb. 11, 2017: **Accepted:** Mar. 28, 2017
***Correspondence Dr. Rajdeep Das.**
Department of Oral Pathology, Haldia institute of Dental Sciences and Research, Haldia, West Bengal, India.
Email: Not Disclosed

pISSN 2278-0076
eISSN 2321-9270

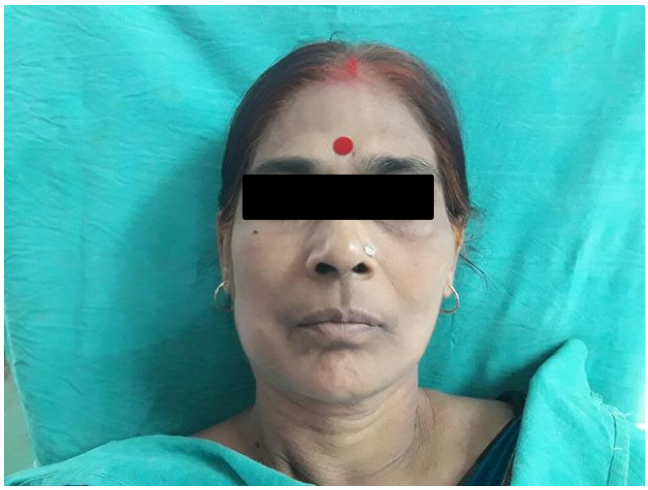

Fig 1: Swelling in right lower back jaw region.

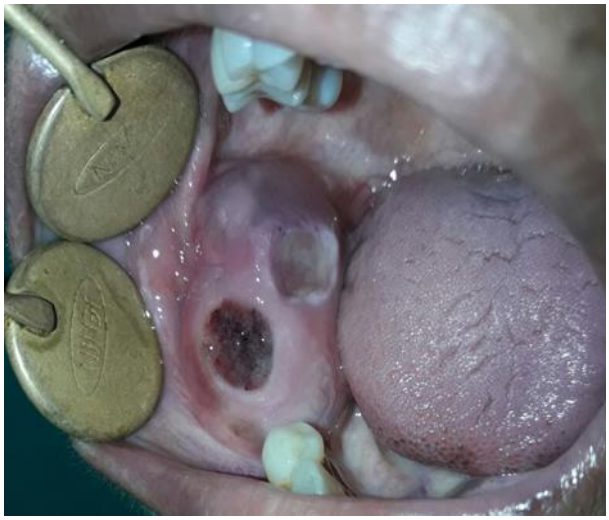

Fig 2: Tilted 46and 47.

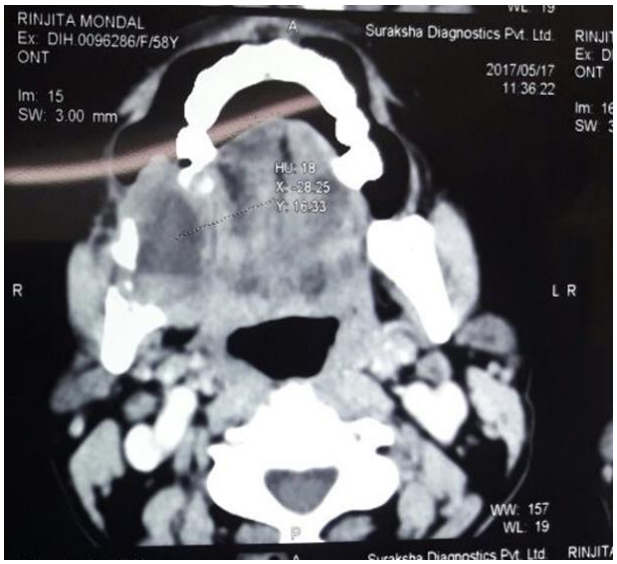

Fig 3: CT scan showing buccolingual expansion of cortical plates of mandible.

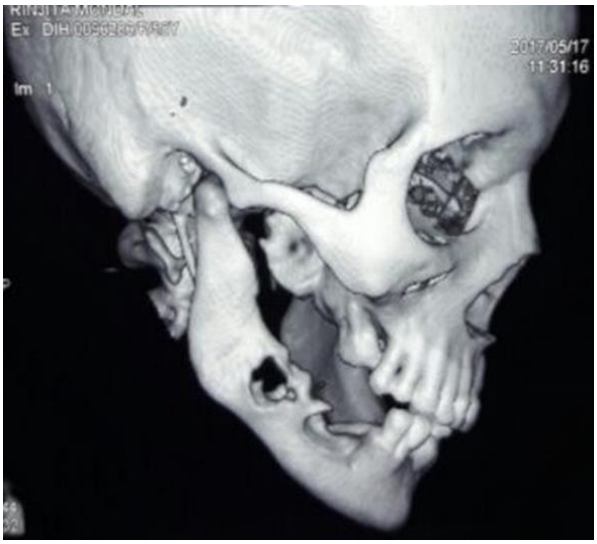

Fig 4: 3D CT showing buccal cortical plate perforation.

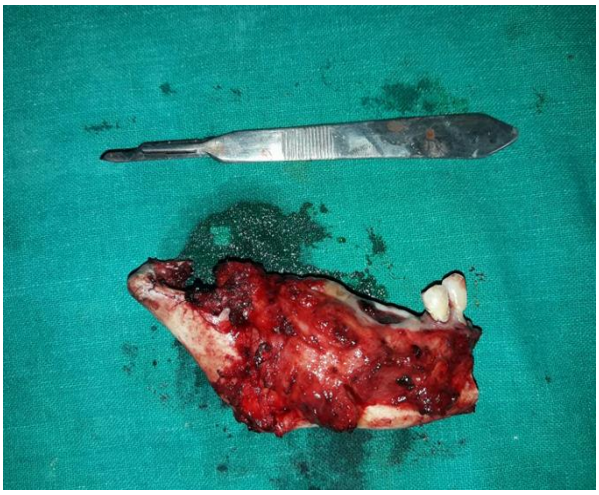

Fig 5: Resected Specimen.

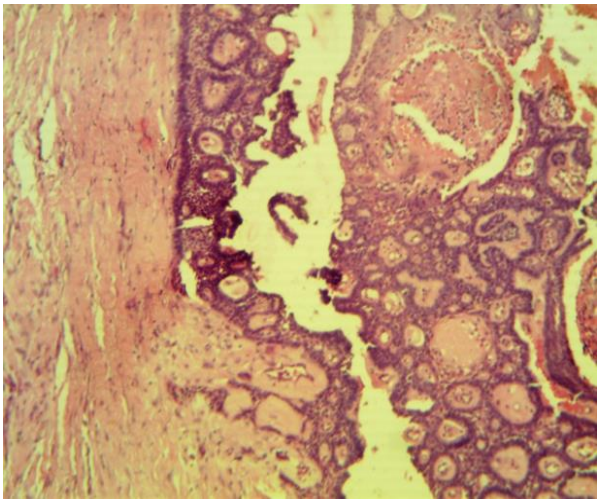

Fig 6: Lining epithelium showing variable thickness.

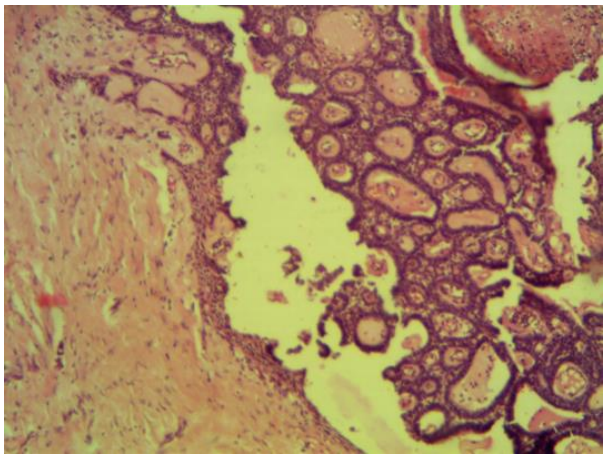

Fig 7: Superficial cell layer showing stellate reticulum like cells.

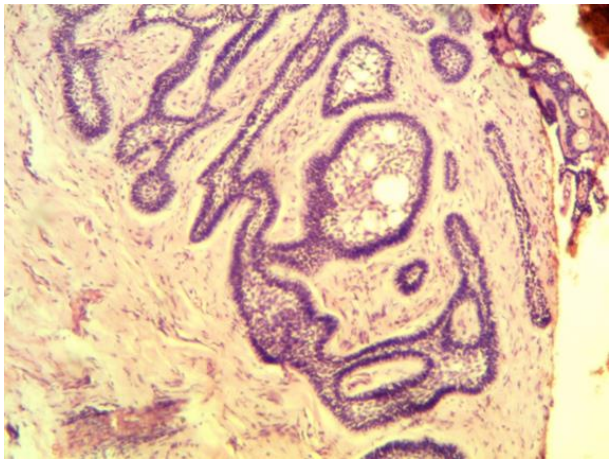

Fig 8: Invading ameloblastomatous islands into the cyst wall.

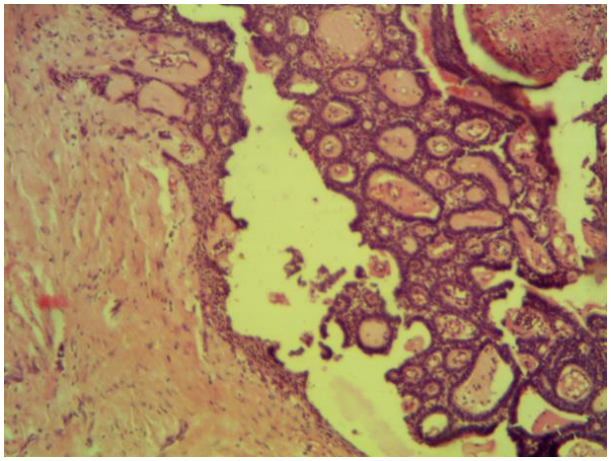

Fig 9: Ameloblastomatous proliferations projecting into the cyst lumen.

in some fields(fig.8). Ameloblastomatous proliferations were found to be projecting into the cystic lumen at few places(fig.9). Based on these findings a report of unicystic ameloblastoma with mural and intraluminal proliferation was given.

Based upon the histopathologic and clinical scenario a hemimandibulectomy of the affected side under general anesthesia was performed. Patient could not afford microvascular reconstruction thus a temporary reconstruction measure with titanium reconstruction plate was attempted. Primary closure was done layer wise. The patients stay was uneventful and is recalled periodically for evaluation.

DISCUSSION

The ameloblastoma is a true neoplasm of enamel organ type tissue which does not undergo differentiation to the point of enamel formation[7]. It arise from either neoplastic transformation of odontogenic cyst epithelium or from residual epithelial rests left over from the formation of teeth, such as remnants of the enamel organ (reduced enamel epithelium) found over the crown of an unerupted tooth, remnants of Hertwig's epithelial root sheath (rests of Malassez) in the periodontal ligament or remnants of the dental lamina (rests of Serres)[8][9].It has been emphasized by clinicians all around the globe that ameloblastoma is benign in nature with local aggressiveness. According to the World Health Organisation, ameloblastomas are classified into following types; conventional, unicystic peripheral and desmoplastic type[10]. Of all the documented cases 20% can be found in the maxillary region and is of particular importance because of their close proximity to vital structures, making it difficult for the surgeon to obtain a clear margin. Out of the remainig 80% of the cases occuring in mandible, majority can be found in ascending ramus and molar area followed by premolar and anterior jaw bone area[11]. The usual age group of the affected patients is found to lie between fourth and fifth decade of life, whereas the unicystic variant is found to be affecting the patients in the age group of second and third decade of life[12][13][14].Clinical symptom usually seen is swelling of jaw bone of variable dimension. Radiological features of ameloblastoma divides this lesion into three categories having distinct features, the most common being the multilocular pattern showing cystic spaces which are either in group or

separated by bony septa exhibiting a soap bubble appearance. The second and third form being honey comb and unicystic appearance with well demarcated border respectively[15].The term unicystic was coined by Robinson and Martinez in the year 1977 and is justified by the fact that both macroscopic and microscopic appearance of the lesion as this type of lesion is having a well defined single cystic sac lined by odontogenic epithelium[16]. Whether unicystic ameloblastoma originated de novo as a neoplasm or whether it is neoplastic transformation of non neoplastic cyst epithelium is matter of long standing debate.They are characterized by slow growth rate and relativey local aggressiveness[17]. The early ameloblastic changes within the cyst wall were first described by Vickers and Gorlin in 1970, and their identification criterion included a cyst lined by ameloblastic epithelium with a tall columnar basal layer,subnuclear vacuoles, reverse polarity of hyperchromatic nucleus and a thin layer of oedematous , degenerating stellate reticulum like cells on the surface[18]. According to the classification given by Ackermann based on clinicopathologic study of 57 cases three histologic types can be identified which are: 1) luminal unicystic ameloblastoma (tumor confined to the luminal surface of the cyst), 2)intraluminal/plexiform unicystic ameloblastoma (nodular proliferation into the lumen without infiltration of the tumour cells into the connective tissue wall), 3) mural unicystic ameloblastoma(invasive islands of ameloblastomatous epithelium in the connective tissue wall not involving the entire epithelium)[19]. Later the classification was modified by Philipsen and Reichart which is: subgroup 1: luminal unicystic ameloblastoma, subgroup1.2: luminal and intraluminal, subgroup 1.2.3: luminal, intraluminal, intramural, subgroup1.3: luminal and intra mural[20].When compared to solid and multicystic ameloblastomas , unicystic ameloblastomas are believed to be less aggressive and respond well to conservative management including enucleation, curettage and marsupialization. More aggressive surgical interventions such as resection should be deferred until recurrence occurs. Unicystic ameloblastoma has a strong tendency to recur particularly when the ameloblastic component penetrates the adjacent tissue structure. For effective treatment of this lesion proper diagnosis is of paramount importance. Among various diagnostic tools, histopathology plays a vital role in proper diagnosis of the lesion. The next tool for diagnosis is molecular markers which may be helpful by identifying cell surface markers like proliferating cell nuclear antigen, Ki67 and calretinin present in ameloblastomatous lesion. Radiographs can also be a helpful tool for providing early detection of the lesion, particularly CT over other radiographs. It has proven its superiority over other radiographs in detection and delineation of intraosseous lesions and their impact on adjacent tissues. Based on these diagnostic aids proper treatment planning can be achieved whether conservative or radical surgical procedure.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

REFERENCES

1. Sciubba JJ, Fantasia JA, Kahn LB, Editors, Benign odontogenic tumors. In atlas of tumor pathology – tumors and cysts of jaw. Washington, D.C :AFIP;1999.p.71-85.

2. Robinson L, Martinez MG. Unicystic ameloblastoma: A prognostically distinct entity. Cancer. 1977; 40:2278-85.

3. Brazis PW, Miller NR, Lee AG, Holliday MJ. Neuro-ophthalmologic Aspects of Ameloblastoma. Skull Base Surg. 1995;5:233–44.

4. Shear M, Singh S. Age-standardized incidence rates of ameloblastoma and dentigerous cyst on the Witwatersrand, South Africa. Community Dent Oral Epidemiol. 1978;6:195–9.

5. Krishnapillai R, Angadi PV. A clinical, radiographic, and histologic review of 73 cases of ameloblastoma in an Indian population. Quintessence Int. 2010;41:e90–100.

6. Kramer IR, Pindborg JJ, Shear M. 2nd ed. Berlin: Springer-Verlag; 1992. Histological Typing of Odontogenic Tumours. WHO International Histological Classification of Tumours; pp. 11–14.

7. Ritchie A (1990) Boyd's text book of pathology, 9th edn. Lea and Febiger, UK

8. Bhasker S (1981) Synopsis of oral pathlogy, 6th edn. The CV Mosby, USA

9. Reichart PA, Philipsen HP, Sonner S (1995) Ameloblastoma: biological profile of 3677 cases. Eur J Cancer B Oral Oncol 31B(2):86–99

10. Wenig BM (2007) Atlas of head and neck pathology, 2nd edn.Elsevier Saunders

11. Philipsen HP, Reichart PA. Unicystic ameloblastoma. A review of 193 cases from the literature. Oral Oncol 1998;34:317e25.

12. Mendenhall WM, Werning JW, Fernandes R, Malyapa RS, Mendenhall NP. Ameloblastoma. Am J Clin Oncol. 2007;30:645–8. [PubMed: 18091060]

13. Dunsche A, Babendererde O, Lüttges J, Springer IN. Dentigerous cyst versus unicystic ameloblastoma-differential diagnosis in routine histology. J Oral Pathol Med 2003 Sep 32(8)486:491.

14. Tsaknis PJ, Nelson JF. The maxillary ameloblastoma: An analysis of 24 cases. J Oral Surg. 1980; 38:336-42.

15. Robinson L , Martinez M.J. Unicystic Ameloblastoma .A prognostically distinct entity. Cancer 1977;40:2278-85.

16. Ricci, et al.Ameloblastoma and reviewe of literature.contemporary clinical dentistry ,septembeer 2012,volume 3, supplement 2.

17. Vickers RA, Gorlin RJ (1970) Ameloblastoma: delineation of early histopathologic features of neoplasia. Cancer 26, 699-710.

18. Ackermann GL, Altini M, Shear M. The unicystic ameloblastoma: A clinicopathological study of 57 cases. J Oral Pathol 1988;17:541-6.

19. Philipsen HP, Reichert PA, Unicystic Ameloblastoma.A review of 193 cases from the literature .Oral Oncol 1998;34;317-25.