CASE REPORT

An Incidental Radiologic Finding of Unicystic Ameloblastoma: A Diagnostic Dilemma

Rishikesh Kumar, Prasanna Kumar D, Dhaya Ann Varghese, Mahabaleshwara C H, Alpha Mary Mathew, Jalashri Shamanewadi

Abstract

Introduction: Benign odontogenic tumors are generally clinically asymptomatic and grows to considerable size before the actual signs and symptoms develop. Sometimes they may be identified during the radiological investigations. Oral and maxillofacial surgeons or pathologist must be aware that the possibility of coming across an unintentional or incorrectly diagnosed Unicystic ameloblastoma. This variant of ameloblastoma is those type of ameloblastoma which have comparable clinical, radiographic and gross features of odontogenic cysts. Therefore, the operating surgeon may be faced with a difficult decision between conservative management or radical approach.

Clinical Presentation: A case report of incidental finding of Unicystic Ameloblastoma in a 63 year old male patient.

Management and Prognosis: Management was done conservatively by enucleation, peripheral ostectomy, and chemical cauterization, and the patient is being followed up.

Conclusion: Adhering to the strategy of conservative management and routine follow-up resulted in a reliable outcome.

Keywords: Ameloblastoma, Odontogenic Keratocyst, Benign, Cauterization

Oral and Maxillofacial Pathology Journal (2023): ***https://www.ompj.org/archives***

Introduction

Ameloblastoma refers to a benign odontogenic tumor of epithelial origin and is locally aggressive with unlimited growth capacity.[1] The tissues involved are most often those that give rise to the teeth so that ameloblastoma may cause facial distortion. Unicystic ameloblastoma has a well-defined, corticated border often associated with an impacted tooth or adjacent root resorption. Macrocystic changes of tumor island in the HPE sample can aid in differentiating ameloblastoma from other cysts.

Among the different types, unicystic ameloblastoma is the less encountered variant.[2] Unicystic ameloblastoma is asymptomatic until gross expansion makes it deleterious. A typical feature of unicystic ameloblastoma is the ability to mimic an odontogenic cyst radiologically and clinically. Though histopathological report can demarcate between the two, chances of error or difficulty in determination of the lining in case of early stages can pose a dilemma to both pathologist and surgeon in deciding a final diagnosis making it strenuous for the surgeon to decide the course of surgical management.

Case Presentation

A 63-year-old male patient reported to the outpatient wing of the department of oral and maxillofacial surgery, KVG Dental College, Sullia with a chief complaint of missing teeth in the upper and lower back tooth region and wanted replacement of teeth with implant prosthesis. Patient underwent extraction of 46 three months back with uneventful healing and gave no history of associated pain or swelling with edentulous region. Patient was known pre-diabetic with lifestyle modification and controlled glycemic levels.

Patient was moderately built, moderately nourished and well oriented to time, place and person. On extra oral examination there were no significant findings or changes noted or palpated. On intra oral examination, missing teeth

Department of Oral and Maxillofacial Surgery, KVG Dental College and Hospital, Sullia, Karnataka, India.

Corresponding Author: Rishikesh Kumar, Department of Oral and Maxillofacial Surgery, KVG Dental College and Hospital, Kurunjibagh, Sullia, Karnataka, Pin-574327, Email: rishishubh550@gmail.com

How to cite the article: Kumar R, Kumar D P, Varghese DA, Mahabaleshwara C H, Mathew AM, Shamanewadi J. An Incidental Radiologic Finding of Unicystic Ameloblastoma: A Diagnostic Dilemma. Oral Maxillofac Pathol J 2023; 14(2). Page number 259-261

Source of Support: Nil

Conflict of Interest: None

were present with 14,16,36,37,46,47 and grossly decayed teeth were present with 26,28. On palpation all the inspectory findings were confirmed and no vestibular tenderness was elicited. Provisionally, it was diagnosed as partially edentulous maxilla and mandible and OPG was advised.

OPG revealed a well-defined radiolucent lesion of size 4x3 cm in size with sclerotic border with edentulous area of 46 and 47. Internal structure had clear radiolucency with locularity seen in mesial aspects of lesion. Lesion appeared to displace inferior alveolar canal downwards, approaching inferior border of mandible. Subsequent to this incidental finding, CBCT was advised which reported, a well-defined radiolucency measuring approximately 39.7x 23.9 mm suggestive of cyst.

FNAC smear showed presence of islands of epithelial cells with prominent nucleus in a background of RBCs. After these investigations, differential diagnosis for this was Odontogenic Keratocyst, Ameloblastoma, Residual Cyst.

To confirm the diagnosis incisional biopsy was done and specimen was sent for histopathological examination which reported as suggestive of odontogenic cyst. As most of these investigations concluded odontogenic cyst, patient was planned for excisional biopsy. Thereafter, enucleation along with peripheral ostectomy and chemical cauterization with modified Carnoy's solution was done under General Anesthesia and the excised sample was sent for final histopathological evaluation which reported as Unicystic ameloblastoma. Patient is being followed up regularly and trimonthly OPG is taken.

Discussion

A unicystic ameloblastoma was first described by Robinson and Martinez in 1977.1 Unilocular ameloblastoma (UA) is a rare variant of ameloblastoma, accounting for about 6%of ameloblastomas.[2] Although it is a subtype of ameloblastomas, it can be distinguished because of its more benign biologic nature and better response to conservative therapy. Most of these cases are difficult to identify and distinguish without any investigations as it often occur asymptomatically.

Unicystic ameloblastomas have a slight male predilection and frequently originate from the posterior mandible.[2] This is a general finding in younger population. Radiographically, the lesions commonly show expansive unilocular radiolucencies with a well-demarcated border. Approximately 50–80% of cases are associated with an impacted or unerupted tooth.[7] Therefore, the clinical and radiographic presentations of unicystic ameloblastoma are sometimes indistinguishable from those of dentigerous cysts.[2,3,4]

In a study to evaluate radiographic features of odontogenic keratocysts (OKCs) and ameloblastomas conducted by Jira Kitisubkanchana et al using radiographs of 100 cases of OKC and 101 cases of ameloblastoma in terms of location, border, shape, association with impacted tooth, tooth displacement, root resorption and bone expansion, they found that unilocular radiolucent lesion with smooth border, no adjacent tooth displacement, no root resorption and causing mild or no bone expansion is suggestive of an OKC rather than an ameloblastoma.

In our case, CBCT revealed apical involvement of mesial root of 48 in relation to its posterior extent and lesion was in close proximity with root apex of 45 anteriorly. There was thinning of buccal and lingual cortex and slight expansion of lingual cortex. On histological examination of incisional biopsy, the lining tissue and bone sample taken, showed mild to moderate chronic inflammatory cell infiltrate predominantly of lymphocytes. One part of section showed stratified epithelium of 6 to 8 cells thick with hyperchromatic basal and suprabasal cells which was reported as suggestive of odontogenic cyst.

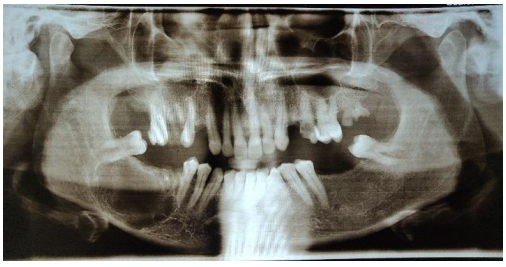


Fig. 1: Well defined radiolucent lesion of size approximately 4x3 cm with a sclerotic border i.r.t 46 and 47. Lesion also appears to displace inferior alveolar canal downwards.

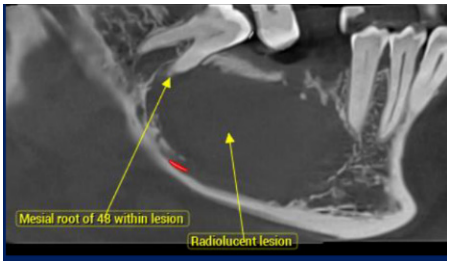


Fig. 2: BCT (Sagittal section) - Well-defined radiolucent lesion measuring 39.7x23.9 cm with involvement of mesial root of 48 within the lesion.

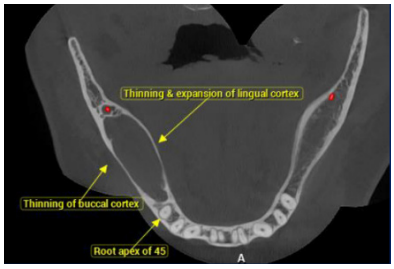


Fig. 3: Pre-Operative CBCT (Axial section)- thinning of buccal and lingual cortical plates and slight expansion of lingual cortex

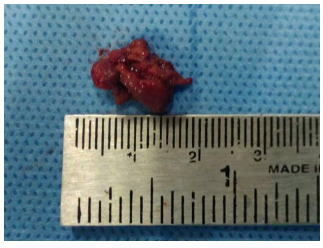
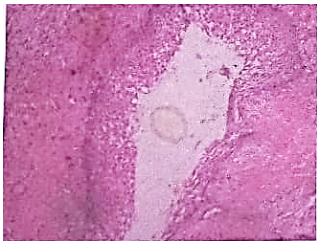


Fig. 4: Gross specimen post enucleation



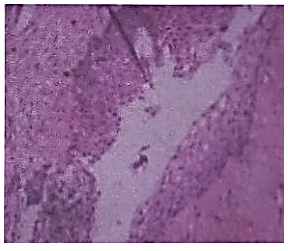


Fig. 5 & 6: Unicystic Ameloblastoma H &E --X

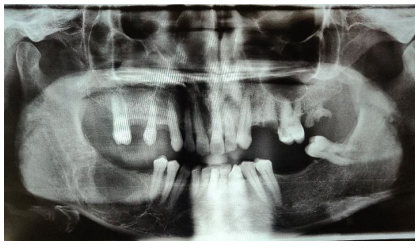


Fig. 7: Post operative OPG - Bone formation and intact inferior border of mandible

Therefore, treatment protocol also followed conservative management with enucleation, peripheral ostectomy and chemical cauterization with Modified Carnoy's Solution. Use of Carnoy's solution has been relatively contraindicated in recent studies due to carcinogenic action of chloroform. Final biopsy of the resected lining histopathologic appearance showed large cystic cavity lined by epithelium of varying thickness exhibiting hyper chromatic, palisading and polarized columnar basal cells. Supra basal cells are loosely textured and non-cohesive resembling like stellate reticulum and cytoplasmic vacuolization in few areas. There was presence of epithelial edema and connective tissue wall consisted of dense bundles of collagen fibers with spindle shaped fibroblasts and in few areas' juxta-epithelial hyalinization. Moderate chronic inflammatory cell infiltrate was also present. Therefore, the final diagnosis post-surgery concluded to be Unicystic Ameloblastoma. Now the dilemma occurs about further management, if conservative management was sufficient or do we have to go for a second surgery for resection. Most authors find no direct relationship between various histological types of the ameloblastoma and the clinical behaviour.[5]

Radical surgery often means that the patients have serious complications including facial deformity, masticatory dysfunction, and abnormal jaw movement.[6] In case of Ameloblastoma, as per literature, most common treatment protocol is resection of adjacent bone involved, but in case of unicystic ameloblastoma, this treatment protocol is still controversial as there are studies that have proved management of unicystic ameloblastoma to be treated primarly like an odontogenic cyst with conservative management and regular follow up. The biologic behavior of this variant tends to be less invasive than multilocular ameloblastomas. They respond more favourably to conservative surgery than do solid or multicystic ameloblastomas.[3,7,8,9,10,11]

A case report by Natália Galvão Garcia et al on Management of Unicystic Ameloblastoma with Mural Proliferation by Conservative Treatment, reported a case of unicystic ameloblastoma that occurred in the right posterior mandible of 19-year-old girl, which was enucleated and did not recur after 12-month follow-up.[12]

Similarly, another case report on Unicystic Ameloblastoma by Ming-HsuanHsu et al reported that conservative interventions are generally preferred for unicystic ameloblastomas in the mandible but are not suggested for those in the maxilla, because of the spongy osteoarchitecture of the maxilla which facilitates spread of the tumor.[12]

It is suggested that a relatively conservative therapies can initially be applied for unicystic ameloblastomas, with more-aggressive approaches being reserved for later recurrence and long-term follow-up is mandatory for unicystic ameloblastomas since recurrence may take place years after removal.[13] Less invasive therapeutic methods can produce beneficial treatments, particularly when combined with the clinical and surgical considerations, in light of the significant fundamental principles in the current research.[14] This approach can only be suggested where it is possible to follow the patient for an extended period of time. In this case, trimonthly OPG is taken for close follow up to observe for healing and any signs of expansion or recurrence. Follow up protocol formulated is trimonthly OPG for a year followed by yearly long term follow up for better prognosis.

Conclusion

This case discussed emphasizes the diagnostic dilemma in a case of unicystic ameloblastoma as it mimics as odontogenic cyst clinically and radiologically. Even though the final HPE report determined that our case was unicystic ameloblastoma, adhering to the strategy of conservative management and routine follow-up resulted in a reliable outcome.

References

1. Robinson L, Martinez MG. Unicystic ameloblastoma. A prognostically distinct entity. Cancer. 1977 Nov;40(5):2278-85.
2. Ramesh RS, Manjunath S, Ustad TH, Pais S, Shivakumar K. Unicystic ameloblastoma of the mandible-an unusual case report and review of literature. Head & neck oncology. 2010 Dec;2(1):1-5.
3. Philipsen HP, Reichart PA. Unicystic ameloblastoma. A review of 193 cases from the literature. Oral Oncology. 1998 Sep 1;34(5):317-25.
4. Bisinelli JC, Ioshii S, Retamoso LB, Moysés ST, Moysés SJ, Tanaka OM. Conservative treatment of unicystic ameloblastoma. American journal of orthodontics and dentofacial orthopedics. 2010 Mar 1;137(3):396-400.
5. Li TJ, Wu YT, Yu SF, Yu GY. Unicystic ameloblastoma: a clinicopathologic study of 33 Chinese patients. The American journal of surgical pathology. 2000 Oct 1;24(10):1385-92.
6. Cernea P, Guilbert F, Rouchon C, Brocheriou C, Payen J. Les récidives des améloblastomes des maxillaires. Etude statistique, indications thérapeutiques. Rev Stomatol (Paris). 1974;75:18.
7. Neville BW, Dam DD, Allen CM, Bouquot JE. Patologia oral maxilofacial. Rio de Janeiro, Brazil: Ed. Guanabara Koogan;2004. p. 499-507
8. Iordanidis S, Makos C, Dimitrakopoulos J, Kariki H. Ameloblastoma of the maxilla. Case report. Aust Dent J 1999;44:51-5.
9. Isacsson G, Andersson L, Forsslund H, Bodin I, Thomsson M. Diagnosis and tretatment of the unicystic ameloblastoma. Int J Oral Maxillofac Surg 1986;15:759-64.
10. Economopoulou P, Sotiriadou S. An unusual tumor of the mandible with features of unicystic ameloblastoma and ameloblastic fibroma. J Oral Maxillofac Surg 1998;56:1196-200.
11. Dandriyal R, Gupta A, Pant S, Baweja HH. Surgical management of ameloblastoma: Conservative or radical approach. National journal of maxillofacial surgery. 2011 Jan;2(1):22.
12. Garcia NG, Oliveira DT, Rodrigues MT. Unicystic ameloblastoma with mural proliferation managed by conservative treatment. Case reports in pathology. 2016 Jan 1;2016.
13. G. Isacsson, L. Andersson, H. Forsslund, I. Bodin, M. Thomsson Diagnosis and treatment of the unicystic ameloblastoma Int J Oral Maxillofac Surg, 15 (1986), pp. 759-764)
14. Queiroz SBF, Amorim RFB, Godoy GP, Freita RA. Tratamentoconservadorem um caso de ameloblastoma so´lido: novosconceitos e abordagensterapeˆuticas. Rev Bras Patol Oral 2002;1:39-46
15. (Li TJ, Kitano M, Arimura K, Sugihara K. Recurrence of unicystic ameloblastoma: A case report and review of the literature. Archives of pathology & laboratory medicine. 1998 Apr 1;122(4):371.)
16. Bisinelli JC, Ioshii S, Retamoso LB, Moysés ST, Moysés SJ, Tanaka OM. Conservative treatment of unicystic ameloblastoma. American journal of orthodontics and dentofacial orthopedics. 2010 Mar 1;137(3):396-400.