

Unicystic ameloblastoma in a 17 year old female – Case report and review of literature

This article was published in the following Scient Open Access Journal:

Journal of Dental and Oral Health

Received October 06, 2019; Accepted October 18, 2019; Published October 21, 2019

Iyogun CA*, Olaitan AA and Sule AA

¹University of Port Harcourt Teaching Hospital, Port Harcourt, Nigeria

²Department of Dental Surgery, Ahmadu Bello University Hospital, Kaduna, Nigeria

³Department of Pathology, Bayero University Kano/ Aminu Kano Teaching Hospital, Kano, Nigeria

Abstract

Introduction: Unicystic ameloblastoma is an infrequent form of ameloblastoma and refers to cystic lesion that show clinic-radiologic features of an odontogenic cyst but histologically reveal a typical ameloblastomatous epithelium lining part of the cystic cavity. It often pose a pose a significant diagnostic and therapeutic challenges. Most of the cases involve the posterior region of the mandible. There are few published cases of unicystic ameloblastoma in our setting, hence this report.

Case report: A 17-year old female presented with four years history of painless frontal lower jaw swelling. There was no history of paraesthesia, bleeding, discharge of fluid or pus from this gradually increasing swelling. Physical examination revealed a well circumscribed swelling in the anterior mandible extending from first premolar on the right to the first premolar on the left mandible. There was egg crackling sensation on palpation of the buccal cortical plate. All teeth were firm except the lower right central incisor which showed grade one mobility. Pulp testing done showed that the teeth were viable. Xray showed a well circumscribed radioluscent lesion with cortical rimming suspected to be a dentigerous cyst. Results of routine baseline investigations were essentially normal. The lesion was enucleated via intraoral buccal flap approach and histology confirmed unicystic ameloblastoma.

Conclusion: Unicystic ameloblastoma involving anterior mandible is rare, meticulous observation, high index of suspicion, radiological and histopathological features are essential in making diagnosis.

Keywords: Unicystic ameloblastoma, Mandible, Histology

Introduction

Ameloblastoma is a benign, slowly growing, locally invasive, epithelial odontogenic tumour of the jaws with a high rate of recurrence if not completely excised. It is classified into solid/multicystic, extraosseous/peripheral, desmoplastic and unicystic types [1]. Unicystic ameloblastoma is an infrequent variant and accounts for 5 – 15% of all ameloblastomas [2]. The mean ages are 16 and 35 years for unerupted and erupted tooth respectively [1]. Vast majority of cases present as painless swelling with facial asymmetry [3]. Radiologically, it presents as a well corticated unilocular radiolucency [4]. The unicystic ameloblastoma is further subdivided into two basic histopathological patterns, the luminal and mural [1]. The treatment depend on clinico-pathological factors such as the size, anatomical site and histologic variant [5]. There is paucity of reports on unicystic ameloblastoma in our setting. We are presenting a case of unicystic ameloblastoma in a 17-year old female.

Case report

A 17-year old female attended the Maxillofacial surgery outpatient clinic with four years history of painless frontal lower jaw swelling. There was no history of paraesthesia, bleeding, discharge of fluid or pus from this gradually increasing swelling. Past dental history was not contributory as the patient had never visited a dental clinic previously. So also the past medical history. Physical examination revealed a well circumscribed swelling in the anterior mandible extending from first premolar on the right to the first premolar on the left mandible (Figure 1). The swelling measured 6cm in diameter with cortical expansion mainly of the buccal plate. The mucosa surface was pink without evidence of ulceration. There was egg crackling sensation on palpation of the buccal cortical plate. All teeth were firm except the lower right central incisor which showed

*Corresponding Author: Cornelius Iyogun, University of Port Harcourt Teaching Hospital, Port Harcourt, Nigeria

grade one mobility. A golden-yellowish fluid with shimmering effect was aspirated from the lesion. Other systemic examinations were unremarkable. Xrays were requested for, which included right/left oblique laterals, occlusal and PA views. It demonstrated a well circumscribed radiolucent lesion with cortical rimming, lamina dura of the affected teeth were intact except for the root of the lower first incisor that was slightly amputated (Figure 1). Results of routine baseline investigations were essentially normal. Clinical diagnosis of dentigerous cyst, /ameloblastoma was made.

The patient was counseled and had enucleation via intraoral buccal flap approach. Portions of the lingual plates overhanging were nibbled to prevent necrosis and subsequent osteomyelitis. Post operative recovery was uneventful. The enucleated lesion was submitted for histopathological examination and revealed cyst wall lined by ameloblasts exhibiting nuclei palisading, reverse polarity and subnuclear vacuoles. Foci of stellate reticulum like cells were noted and diagnosis of unicystic ameloblastoma was made (Figures 2,3). Showed the histology of unicystic ameloblastoma. The patient showed remarkable improvement without any evidence of recurrence for over four years of follow-up visits.

Discussion

Unicystic ameloblastoma is a rare variant of ameloblastoma, presenting as a cyst. It accounts for 5 - 15% of all ameloblastomas and there are many debates concerning the origin of unicystic



Figure 1: occlusal Xray showing a radiolucent lesion of lower anterior mandible (UNICYSTIC AMELOBLASTOMA)

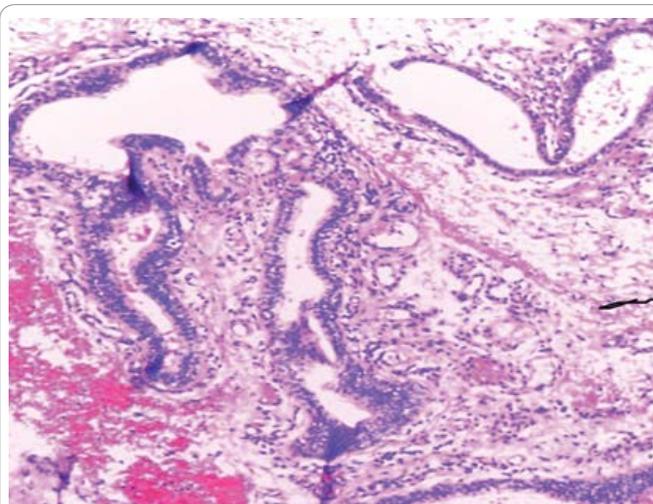


Figure 2 : Unicystic ameloblastoma (H&E x10)

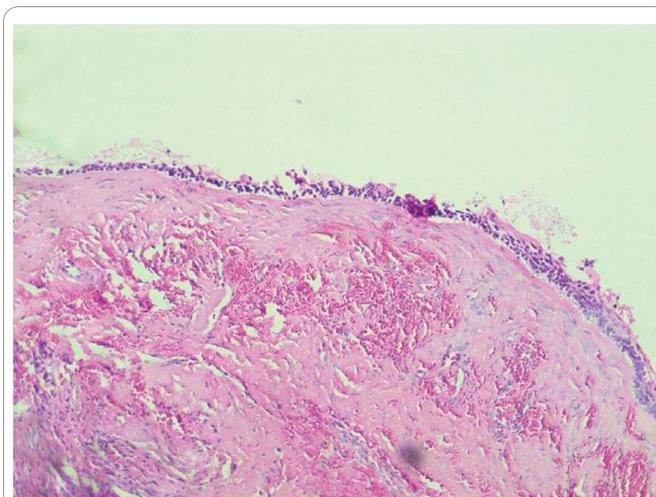


Figure 3: Photomicrograph of lining of unicystic ameloblastoma

ameloblastoma whether it arises de novo or from non-neoplastic cyst epithelium [3]. Leider et al, proposed three pathogenic mechanisms for the evolution of unicystic ameloblastoma: reduced enamel epithelium, from dentigerous cyst and due to cystic degeneration of solid ameloblastoma [6]. It often involves an impacted tooth and the focal area of the cystic tumour lining is often composed of a non specific, thin epithelium that mimics the dentigerous cyst lining [7]. The origin of the index case is not clear.

Unicystic ameloblastoma tends to affect young patients, with about half of cases occurring in the second decade of life [8]. The index case is a 17-year old female. Mandible is the common site of involvement in more than 90% of cases, usually the posterior region [1]. This is in contrast with the present case which was found in the anterior mandible. Majority of cases are asymptomatic, sometimes presenting as painless swelling of the jaws as seen in our case. Atimes, it may grow and causes mucosa ulceration. Small cysts are usually detected by routine radiographs or due to local effects of the lesion

Radiological examination is vital in making diagnosis and planning for surgery. It presents as a well corticated unilocular radiolucency. Root resorption can be seen.4 The index case showed a well circumscribed radiolucent lesion with cortical rimming suspected to be a dentigerous cyst. Dentigerous cysts are mostly seen in panoramic radiographies as unilocular, well-defined radiolucent areas that show cortication. In addition, dentigerous cyst may resemble odontogenic keratocyst and ameloblastic fibroma on plain radiography. Odontogenic keratocysts are not always pericoronal. They cause less expansion and less root resorption. Ameloblastic fibromas are expansive unilocular or multilocular radiolucencies that are well-defined, corticated border and mostly they do not have tooth structure or calcification within the lesion. In many cases, they are associated with the crown of an unerupted tooth. Computed tomography (CT) provide important information for the differential diagnosis [9].

The definitive diagnosis is based on histopathological examination. It is divided into luminal and mural type. In luminal variant, the tumour is confined to the luminal surface. The

present case show no evidence of tumour infiltration within the fibrous wall. Whereas in the mural variant, there are invasive islands of ameloblastomatous epithelium within the fibrous wall. Dentigerous cyst are usually lined by stratified squamous epithelium [10]. Average interval of recurrence is 7 years. Recurrence is also related to histologic subtypes of unicystic ameloblastoma, with those invading the fibrous wall having a rate of 35.7%, but others only 6.7% [8]. The index patient had no evidence of recurrence for over four years of follow-up visit. Long-term follow-up is necessary because of occasional cases of delay recurrence of unicystic ameloblastoma [11].

Conclusion

Unicystic ameloblastoma involving anterior mandible is rare, meticulous observation, high index of suspicion, radiological and histopathological features are essential in making challenging diagnosis.

Conflict of interest

None declared

References

1. Barnes L, Everton JW, Reichart P, Sidransky D. World health organization classification of tumours: Head and neck tumours. Lyon, France, IARC Press. 2005;284-302.
2. Gupta N, Saxena S, Rathod VC, Aggarwal P. Unicystic ameloblastoma of the mandible. *J Oral Maxillofac Pathol.* 2011;15:228-231.
3. Ackermann GL, Altini M, Shear M. The unicystic ameloblastoma: A clinicopathological study of 57 cases. *J Oral Pathol.* 1988;17:541-546.
4. Gungum S, Hosgoren B. Clinical and radiologic behaviour of ameloblastoma in 4 cases. *J Can Dent Assoc.* 2005;71:481-484.
5. Williams TP. Management of ameloblastoma: A changing perspective. *J Oral Maxillofac Surg.* 1993;51:1064-1070.
6. Leider AS, Eversole LR, Barkin ME. Cystic ameloblastoma. *Oral Surg Oral Med Oral Pathol.* 1985;60:624-630.
7. Gardner DG. A pathologist's approach to the treatment of ameloblastoma. *J Oral Maxillofac Surg.* 1984;42:161-166.
8. Li TJ, Wu YT, Yu SF, Yu GY. Unicystic ameloblastoma: A clinicopathologic study of 33 Chinese patients. *Am J Surg Pathol.* 2000;24:1385-1392.
9. Zerrin E, Husniye DK, Peruze C. Dentigerous cysts of the jaws: Clinical and radiological findings of 18 cases. *J Oral Maxillofac Radiol.* 2014;2:77-81.
10. Adebisi KE, Ugboko VI, Esan GO, Ndukwe KC, Oginni FO. Clinicopathological analysis of histological variants of ameloblastoma in a suburban Nigerian population. *Head & Face medicine.* 2006;24(2):42.
11. Olaitan AA, Adekeye EO. Unicystic ameloblastoma of the mandible: A long-term follow-up. *J Oral Maxillofac Surg.* 1997;55:345-350.