Case Report

DOI: 10.21276/APALM.3122

Acanthomatous Ameloblastoma: A Rare Presentation

R. N. Hathila, Pinal C. Shah, Archana V. Patel, Kuntal Patel* and Nilam J. Tejani

Department of Pathology, GMC Surat

ABSTRACT

Ameloblastoma is a slow growing, locally aggressive neoplasm of enamel organ type tissue with a high propensity for recurrence. They occur either in maxilla or mandible at nearly any age but most frequently are discovered as a painless expansion in the mandible of patients in their 20s – 40s with equal frequencies in male and female. Histopathologically the follicular and plexiform patterns are the most common. When extensive metaplasia associated with keratin formation occurs in central portions of the epithelial islands of follicular ameloblastoma, the term Acanthomatous is applied. Here we present a case of Acanthomatous ameloblastoma in a 40 years old female patient.

Keywords: *Acanthomatous, Ameloblastoma, Maxilla, Mandible, Neoplasm*

Introduction

Ameloblastoma is a rare odontogenic tumor of jaw which is benign but locally aggressive in nature. Broca in 1863 was the first to report as adamantinoma and then Churchill in 1934 suggested an alternate name called "ameloblastoma ". The world health organization (WHO) (1991) defined ameloblastoma as a benign but locally aggressive tumor with a high tendency to recur, consisting of proliferating odontogenic epithelium lying in a fibrous stroma. Ameloblastoma is most commonly seen in the posterior mandible but may also arise in the maxilla and anterior aspect of jaw. The radiographic appearance ranged from a unilocular to a multilocular radiolucency, soap bubble and honey comb appearance. There are four different macroscopic subtypes- Solid or multicystic, unicystic, desmoplastic and peripheral. This classification may have a prognostic value.

There are six histologic subtypes of ameloblastoma: follicular, plexiform, acanthomatous, granular, basal cell and desmoplastic. They can be found combined or isolated and that are not related to prognosis of the tumor. Ameloblastoma is rarely metastasizing.

Case Report

A 40 years old female presented to our hospital with a swelling over right side of face since 3 years of duration, which was slowly increasing in size. After right segmental mandibulectomy, we received specimen measuring 8x7x1 cm3 in size along with presence of 6 teeth, brownish in colour. Mandible is replaced by growth. Outer surface is smooth, no areas of haemorrhage or necrosis identified. On serial cutting, multiple whitish fibrotic and occasional cystic areas identified. No areas of haemorrhage/ necrosis seen.

Microscopy revealed nests and islands of epithelium with peripheral palisading of tall columnar cells with reverse polarity (basaloid). The central portion of island shows loose network of reticular cells. Squamous metaplasia with keratin formation is seen in reticular cells. Tumor is limited by fibrous capsule. Marked nuclear atypia, mitosis/ necrosis are not seen in sections examined. Lymphovascular/ neural invasion are not seen.

On the basis of above histological features, diagnosis of ameloblastoma – Acanthomatous type was made.

Discussion

Ameloblastoma accounts for 1% of all tumours of the jaw encountered during 3rd and 5th decade of life. [2] Average age reported is in the third to fifth decade of life which is consistent with our case as the patient in our case was in 4th decade of life. About 80 % of the ameloblastoma occurs in the mandible, out of which 70% are located in the area of molars and ascending ramus, 20% occur in the premolar region and 10% in the areolar region.

Ameloblastoma has been classified in both human and veterinary literature and has been defined as benign, locally invasive and clinically malignant lesions. The potential source of this tumours are the cell rests of the enamel organ, epithelial odontogenic cysts, basal cells of surface, epithelium of the jaws and heterotrophic epithelium in other parts of body.[1]

Radiographic appearance of ameloblastoma can vary according to type of tumour. In early stage the lesion may appear cystic, unilocular and may resemble a downgrowth or a residual cyst. Later it becomes multilocular with internal components separated by radio-opaque septa or trabeculae. The classic radiographic appearance of a mixed

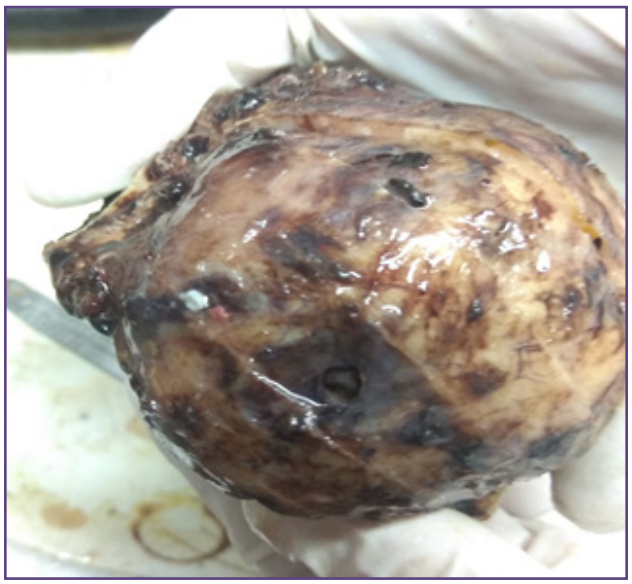

Fig. 1: Outer surface of gross specimen is smooth. Mandible is replaced by growth.

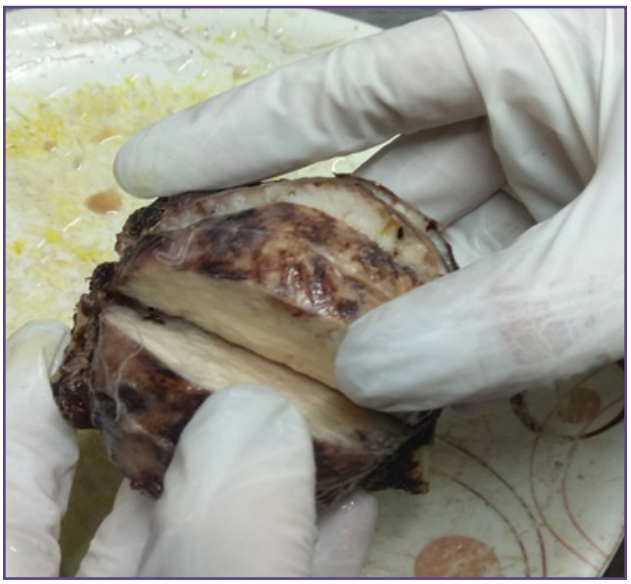

Fig. 2: Cut surface showing multiple whitish fibrotic and occasional cystic areas. No areas of haemorrhage and necrosis.

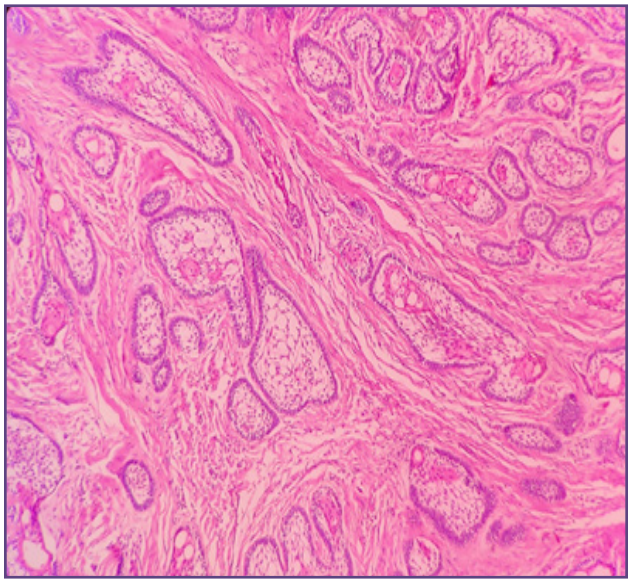

Fig. 3: Nest and island of epithelium with peripheral pallisading of tall columnar cells with reverse polarity (H&E, 10X)

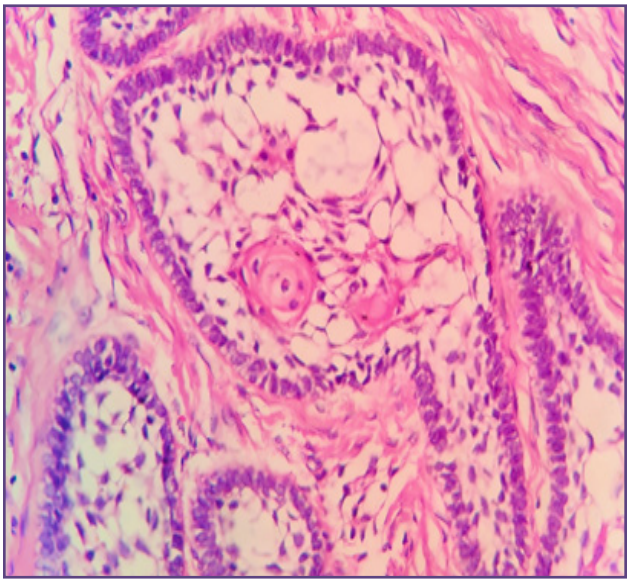

Fig. 4: Central portion of island show loose network of reticular cells. Squamous metaplasia with keratin formation is seen in reticular cells (H&E, 40 X).

cystic or solid form of ameloblastoma is a multilocular radiolucency with a "soap bubble" or "honey comb" appearance. [2]

Microscopically, ameloblastomas are categorised into follicular, plexiform, acanthomatous, basaloid, granular and desmoplastic.[3] out of which follicular is the most prevalent variant while acanthomatous is the rarest type.

Acanthomatous type is benign tumour but is locally aggressive and frequently invades the alveolar bone or recurs after marginal surgical excision. Patients may complain or present with history of a slow growing mass, malocclusion, loose teeth or more rarely paraesthesia and pain however many lesions are detected incidentally on radiographic studies in asymptomatic patients.[4]

Dentigerous cyst is one of the differential and differentiated by absence of superficial keratinization.[3] Osteolytic bony lesions due to metastasis of squamous cell carcinoma to jaw can also be one of the differentials, particularly when

squamous component of acanthomatous ameloblastoma looks atypical. Diligent search for basaloid cells and stellate reticulum can avoid misdiagnosis.[2]

Histopathological examination of acanthomatous Ameloblastoma shows squamous metaplasia of stellate reticulum and the formation of keratin within the tumour islands.[5]

In humans malignant ameloblastomas and ameloblastic carcinomas have been noted to metastasized to the lungs, pleura, orbit, skull and brain.[4] Although intraosseous ameloblastoma is histologically benign and locally aggressive, it only rarely metastasizes.[6]

Although odontogenic tumours have particular histological characteristics, it is not uncommon for them to be misdiagnosed by pathologist who are not familiar with oral pathology.

The treatment of choice is complete surgical resection. If possible, conservative surgery can be used if an assured complete removal can be done.[4] In the present case, surgical resection of the lesion was done.

Conclusion

Ameloblastoma is uncommon benign odontogenic neoplasm that rarely become malignant. To conclude, the definitive diagnosis is based on its histopathological appearance. Documentation and research of such cases may helpful to correlate the prognosis of the lesions.

Acknowledgements

We are thankful to Dr. Mandakini M. Patel (HOD), Dr. R. N. Hathila (Additional professor) and other faculty members and technical staff of department of pathology at Government medical college, Surat for their help and support.

Funding

No source of funding

Competing Interests

Not applicable

References

1. Jeddy N, Jeyapradha T, Ananthalakshmi R, Jeeva S, Saikrishna P, Lakshmipathy P. The molecular and genetic aspects in the pathogenesis and treatment of ameloblastoma. 2013;2(3):157–61.
2. Report C. Case Report Cytological diagnosis of acanthomatous ameloblastoma. 2008;25(2):156–8.
3. Goldblum JR, Lamps LW, McKenney JK, Myers JL. Rosai and Ackerman's Surgical Pathology. 11th ed. Elsevier; 2018. 224–226 p.
4. Singh G, Agarwal R, Kumar V, Passi D. Acanthomatous Ameloblastoma- A Case Report. 2013;5(2):54–8.
5. Malathi N, Giri GV V, Pandyan DA, Suganya R, Thamizhchelvan H. Case Report Acanthomatous Ameloblastoma of Mandible in a Paediatric Patient. 2018;2018.
6. Mills SE. Sternberg's diagnostic surgical pathology. 6th ed. Wolters Kluwer; 2015.
7. Bhargava A, Saigal S, Chalishazar M. Acanthomatous ameloblastoma of mandible. J Dent Sci Res 2011; 2:1-5.
8. Adel K. El-Naggar, John K.C. Chan, Jennifer R. Grandis, Takashi Takata, Pieter J. Slootweg. WHO classification of head and neck tumors 4th edition, 2017. 215-221 p.

***Corresponding author:**
Dr. Kuntal Patel, At post Kherlav, patel faliya, taluka pardi district valsad - 396145
Phone: +91 8511553416
Email: kuntalpatelpk27@gmail.com

Financial or other Competing Interests: None.

Date of Submission : 31/08/2021
Date of Final Revision : 10/12/2021
Date of Acceptance : 29/01/2022
Date of Publication : 28/02/2022