CASE REPORT

Indian Journal of Contemporary Dentistry
Volume 13 No.1, January-June 2025

Anterior Ameloblastoma, Masquerading a Residual Cyst: A Case Report

Shamaila Siddiqui[1*], Malu Pooja Radhesham[2], Vahanwala Sonal P[3], Singh Shivani[4], Soni Sakshi[5], Pooja Prasad[6]

[1,5] Post-graduate Student,department of Oral Medicine and Radiology, Nair Hospital Dental College [2]Asisstant Professor department of Oral Medicine and Radiology,Government Dental College, Aurangabad,[3]Associate Professor department Of Oral Medicine and Radiology, Nair Hospital Dental College,of Oral Medicine and Radiology, Nair Hospital ,[4]MDS Department Of Oral Medicine and Radiology, Nair Hospital Dental College [6]Assisstant Professor Department Of Oral And Maxillofacial Pathology,Nair Hospital Dental College

How to cite this article: Siddiqui S, Radhesham MP, Sonal VP, Shivani S, MDS, Sakshi S, Prasad P,. Anterior Ameloblastoma, Masquerading a Residual Cyst -A Case Report. 2025;13(1):7-12.

ABSTRACT

Background: Ameloblastoma is a benign odontogenic tumor of epithelial origin that exhibits a locally aggressive behaviour with a high level of recurrence and multiple factors involved in its molecular pathogenesis. Anterior ameloblastoma is a rare variant that makes up about 10% of all its varieties, presents unique difficulties in diagnosis, especially while differentiating it from other lesions in anterior region. Its vivid range of clinical manifestation further puzzles in clinical diagnosis and can complicate planning of surgeries and thereby affect the prognosis. We highlight one such case which mimics a residual cyst and thus mandates importance of thorough diagnostics.

Case Report: This case report presents a rare occurrence of ameloblastoma in anterior mandible of a 60-year-old patient who reported a swelling in the lower anterior region, which had been present for 5-6 months and was initially noticed two years ago when it was smaller. This case report aims to highlight the unusual location of the tumor, the clinical and radiological findings and the treatment modalities employed.

Conclusion: Promptness in diagnosis and relevant treatment of ameloblastoma is crucial due to its malignant potential and recurrence rate. It underscores the importance of early detection and intervention, which can significantly improve patient prognosis and reduce the risk of recurrence. Further, it emphasizes the need for regular follow-ups to monitor any risk of recurrence or any other complications.

KEYWORDS: Odontogenic cyst ,Odontogenic tumors, Ameloblastoma , Residual cyst , Anterior Mandible

INTRODUCTION

A multitude of benign lesions fare manifested in the oral and maxillofacial region which could be both odontogenic and non-odontogenic origin. They range from ameloblastoma, radicular cysts, dentigerous cysts, central giant cell granuloma, osteomas, and keratocystic odontogenic tumors. Among these, ameloblastoma, is the most prevalent with The pooled incidence rate of ameloblastoma being 0.92 per million person-years.[11] They account for 1% of all oral tumours.[11] It is an

Corresponding author: Shamaila Siddiqui, Post-graduate Student, Department of Oral Medicine and Radiology, Nair Hospital Dental College

Email: shamaila.sd@gmail.com

Submission: Jan 23, 2025 Accepted: Feb 7, 2025

aggressive odontogenic tumor that forms from odontogenic epithelium within a mature fibrous stroma devoid of odontogenic ectomesenchyme [1].Although classified as a benign tumor , it is notorious in nature as chances of recurrence is high. Thus, most relapses (50% and even over 80%) occur during the first 5 years after the primary surgery [2]. The major contributing factor for recurrence seems to be the inadequate initial surgical procedure rather than the histological type[2] .Ameloblastoma is a slow-growing tumor and a metaanalysis calculated the mean specific growth rate of ameloblastoma to be 87.8% per year [12]. However, if left untreated, ameloblastoma can grow to a huge size and pose a risk to the airway.[12]The current mainstay of treatment is wide local excision with appropriate margins and immediate reconstruction. In this case study, we discuss a particular instance of ameloblastoma, detailing its clinical and radiological observations, the chosen treatment approach, and the findings from the histological analysis. We also provide a follow-up on the case.

CASE PRESENTATION

A 60-year-old patient reported to the Department with swelling in right mandibular region persisting for 5-6 months. The patient who was initially asymptomatic, noticed a minor swelling in the jaw two years ago, which progressively enlarged to present state. The patient also reported tooth mobility and exfoliation of permanent canine in the right mandibular anterior region two years back. There was no associated pain, pus discharge, or fever. Past medical history was non-contributory.

CLINICAL FINDINGS

Extra-oral examination revealed a swelling in right lower half of the mandible measuring approximately 4x2cm in size extending from left mento-labial sulcus, crossing the midline and progressing into ascending ramus of mandible on the right side. This swelling on palpation was bony hard, non-tender and without any raise in temperature. The overlying skin appeared smooth and of normal colour.

On intraoral examination, multiple teeth were missing and 44,45 showed Grade II mobility with obliteration of labial and lingual vestibule [Figure 1].

RADIOLOGICAL FINDINGS

The OPG prescribed showed well-defined radiolucency in the right mandible para symphysis region extending anterioposteriorly from the midline till distal aspect of 45 and superioinferiorly from crestal bone to just short of the inferior border of mandible. The inferior border of the lesion was surrounded by a corticated border with the evidence of scalloping resulting in a multilocular appearance in the premolar region. There was no evidence of root resorption in 44,45 as shown in the pre-operative osthopantomogram [Figure II]. Clinicoradiological differential diagnosis of residual cyst, central giant cell granuloma and ameloblastoma was given.

figure I Showing intraoral clinical picture showing buccolingual expansion of the lesion

Figure II: Orthopanthomogram showing well defined multilocular radiolucent lesion extending anterioposteriorly from the midline till distal aspect of 45 and superioinferiorly from crestal bone to just short of the inferior border of mandible. The inferior border of the lesion surrounded by a corticated border with the evidence of scalloping resulting in a multilocular appearance in the premolar region.No evidence of root resorption in 44,45

The patient, after all laboratory investigations was prepared for surgery where first and second premolars were extracted. Given that ameloblastoma was a consideration in the differential diagnosis, an extensive surgery was carried out A mucoperiosteal flap was reflected, exposing the tumor, following negative aspiration, en-bloc resection was done [Figure IV] A reconstruction plate was adapted from 35 to 45 region to align and stabilize the bone fragments with three holes on either side of the cut. Intermaxillary fixation screws were drilled to secure the reconstruction plate to the bone and anchored with stainless streel wire. [Figure V]. The periosteum and muscles were released for resection. Osteotomy cuts were made in the anterior region and 1cm beyond the tumor margin in 35 region, completed through the lingual cortex. The middle cut was completed with an osteotome and mallet, and the specimen was removed. A pre-contoured reconstruction plate was secured with screws.Surgery is the mainstay in the treatment of ameloblastoma which can involve complete removal of the tumor with a negative margin of 15-20 mm for controlling the local invasion.This tumour induces aggressive invasion into the surrounding bone and has a high recurrence rate after surgery [10] therefore en-bloc resection with simultaneous recontruction can address this issue.

The acquired specimen was sent for histopathological examination which revealed a tumor mass with typical follicular pattern with islands of odontogenic epithelium within fibrous stroma. The epithelium consisted of peripheral tall columnar palisading cells with reversal of polarity, resembling ameloblasts like cells and central loosely arranged cells resembling the stellate reticulum. [fig III]. The patient was monitored continuously for one year to detect any potential recurrence of ameloblastoma. [Figure V1]

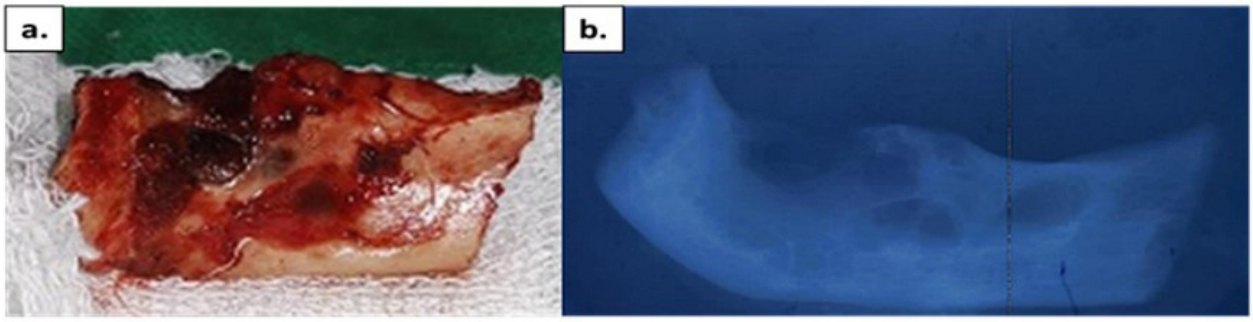


Figure IV: a. Gross specimen of the excised tumor; b. Radiograph of the resected specimen showing typical soap bubble appearance of Ameloblastoma.

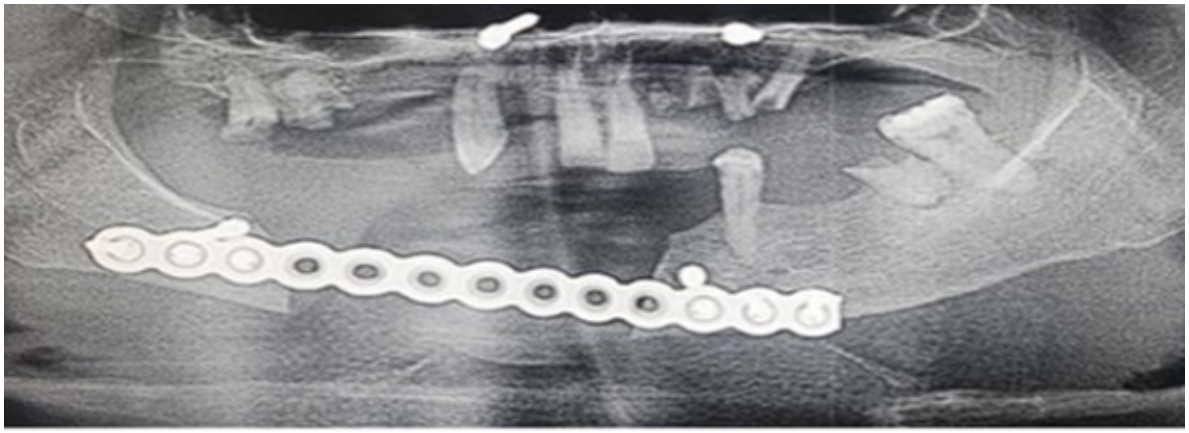


Figure V: OPG taken post-surgery showing secured bone plate extending from mid third quadrant to mid fourth showing the surgical site immediately after surgery

DISCUSSION

Ameloblastoma is a benign, gradually developing, locally invasive tumor of epithelial odontogenic origin appearing in the jaw bones. If not appropriately treated, ameloblastoma has a high potential for recurrence. Review of literature reveals several etiologies hypothesized but have not explicitly been elucidated, with diversifications in the mitogen-activated protein kinase (MAPK), sonic hedgehog (SHH), and WNT/β-catenin pathways being the most common[3]. BRAF V600E gene mutations appeared to be most common in the MAPK pathway. They are commonly identified in mandibular ameloblastomas, while SMO gene mutations are the most common in the non-MAPK pathway and are frequently found in maxillary ameloblastomas. [4]

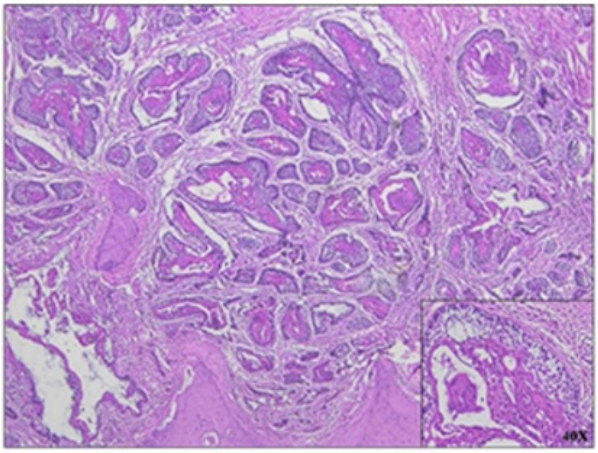


Figure III: Follicular ameloblastoma showing peripheral tall columnar palisaded cells and central stellate reticulum like cells with cystic degeneration and squamous metaplasia.

Radiographically, ameloblastoma appears as a well-defined, unilocular or multilocular, often quite large radiolucency. Root resorption or root divergence may also be observed. Since none of the radiological features are pathognomonic, the definite diagnosis of ameloblastoma is only established by biopsy

In the present case, a large multilocular radiolucency was seen with interrupted borders associated with edentulous alveolar ridge. Also

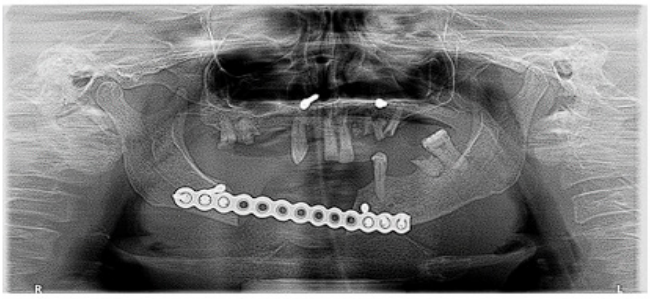


fig VIa: OPG showing 1 month follow up

fig VIb:OPG showing 3 month follow up

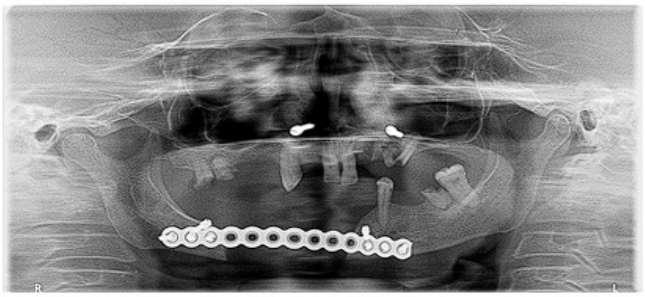
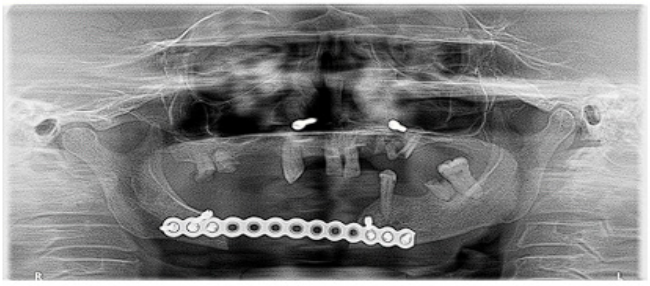


Fig VIc:OPG showing 1 year follow up



no root resorption or divergence was noted. Thus, the radiographic presentation in the present case could be suggestive of residual cyst.

Residual cysts should be high on the list of differential diagnosis when elderly, edentulous patients present with cystic lesions in the jaws compared to dentate patients. These lesions have the ability to destroy bone within the jaws without any symptoms. [7] Clinicopathological features that aid in their diagnosis included long-standing history with slow growing swelling and presence of well-defined, unilocular cystic lesion associated with previously extracted dentition. Moreover, they can mimic more aggressive cysts and tumours on radiographs. odontogenic cysts have the potential to transform into neoplastic lesions.[7]

The radiographic features of Central Giant Cell Granuloma which were seen in this case include its location and multilocularity crossing midline.[9]

Several studies state the epithelial remnants of odontogenic cysts as the main origin of odontogenic cysts transforming into neoplastic lesions, such as ameloblastomas, adenomatoid odontogenic tumours, and even nonodontogenic malignant tumours.However, the frequency of these neoplastic transformations are known to be low [8]. The differentiation of this variant of ameloblastoma from residual cyst arising from edentulous jaw is important since the former requires conservative management while the latter needs aggressive treatment.

One strategy that has the potential to overcome this problem is to use targeted therapy as an adjuvant treatment immediately after surgery. For this strategy to be implemented, the researchers need to find reliable and validated specific biomarkers of ameloblastoma of late. the BRAF V600E mutation has emerged as a predictive, diagnostic, and prognostic biomarker in ameloblastomas. [13]

Targeted therapies which use specific molecules such as genes and proteins for therapeutic reasons, are gaining popularity in treating tumors and malignancies.New molecular medicines that target the underlying oncogenic driver mutations would be a highly desirable addition or alternative to conventional surgery.[14]

CONCLUSION

Knowing the malignant transformation rate and rate of recurrence of ameloblastoma, its precise and prompt diagnosis and treatment is imperative. Hence a clinician should be updated regarding the clinical and radiographic features of ameloblastoma. If variation in clinical and radiographic features of ameloblastoma are noted in the clinical setup, it is advisable that such cases be reported. Upgrading the literature with these variations will enlighten the path to diagnosis and treatment of this odontogenic entity.

It is of great relevance that the radiologist and surgeon discuss and sketch the plan of action prior to surgery so that the lesion is completely resected and removed in toto. This practise enhances good patient care and eliminates the chances of recurrence. Rehabilitation of such patients is important to compensate for loss of function. Regular follow up works both ways for the patient and clinician.it serves as a clinical audit to generate confidence in the patient for follow up.

LIMITATIONS

- This Case studies typically focus on a single patient, which may not represent the larger population.
- Findings from a single case may not be applicable to other cases or broader contexts.

Informed Consent was taken from the patient for using the radiographs, no personal facial image was used.

Source of Funding: None

Ethical clearance: As it is a Case Report ethical clearance is not required.

Conflict of Interest : None

References

1] Effiom OA, Ogundana OM, Akinshipo AO, Akintoye SO. Ameloblastoma: current etiopathological concepts and management. Oral diseases. 2018 Apr;24(3): 307-16.

2] Medina A, Velasco Martinez I, McIntyre B, Chandran R. Ameloblastoma: clinical presentation, multidisciplinary management and outcome. Case Reports in Plastic Surgery and Hand Surgery. 2021 Jan 1;8(1):27-36.

3] Guan P, Wong SF, Lim JQ, Ng CC, Soong PL, Sim CQ, Ong CK, Rajasegaran V, Myint SS, Lee JY, Tan HK. Mutational signatures in mandibular ameloblastoma correlate with smoking. Journal of Dental Research. 2019 Jun;98(6):652-8.

4] Brown NA, Rolland D, McHugh JB, Weigelin HC, Zhao L, Lim MS, Elenitoba-Johnson KS, Betz BL. Activating FGFR2–RAS–BRAF mutations in ameloblastoma. Clinical Cancer Research. 2014 Nov 1;20(21):5517-26.

5] Sweeney RT, McClary AC, Myers BR, Biscocho J, Neahring L, Kwei KA, Qu K, Gong X, Ng T, Jones CD, Varma S. Identification of recurrent SMO and BRAF mutations in ameloblastomas. Nature genetics. 2014 Jul;46(7):722-5.

6]. Heikinheimo K, Kurppa KJ, Elenius K. Novel targets for the treatment of ameloblastoma [Internet]. Vol. 94, Journal of Dental Research. SAGE Publications Inc.; 2015 [cited 2021 Jun 30]. p. 237–40.

7]Titinchi F, Morkel J. Residual cyst of the jaws: A clinico-pathologic study of this seemingly inconspicuous lesion. Plos one. 2020 Dec 17;15(12):e0244250.

8]Kondamari SK, Taneeru S, Guttikonda VR, Masabattula GK. Ameloblastoma arising in the wall of dentigerous cyst: Report of a rare entity. Journal of oral and maxillofacial pathology. 2018 Jan 1;22(Suppl 1):S7-10.

9] Krishnamurthy V, Haridas S, Garud M, Vahanwala S, Nayak CD, Pagare SS. Radicular cyst masquerading as a multilocular radiolucency. Quintessence International. 2013 Jan 1;44(1).

10] Fuchigami T, Ono Y, Kishida S, Nakamura N. Molecular biological findings of ameloblastoma. Japanese Dental Science Review. 2021 Nov 1;57:27-32.

11]Rayamajhi S, Shrestha S, Shakya S, Bhandari S, Twayana AR, Shahi K. Unicystic ameloblastoma of mandible: A case report. JNMA: Journal of the Nepal Medical Association. 2022 Jul;60(251):657.

12]Ghai S. Ameloblastoma: an updated narrative review of an enigmatic tumor. Cureus. 2022 Aug;14(8).

13] Malakar A, Kumar VR, Yadav P, Bhardwaj V, Barua CG, Bhardwaj G. The Role of BRAF Inhibitors in the Management of Ameloblastoma: A Literature Review. Cureus. 2023 Oct;15(10).

14] Nguyen J, Saffari PS, Pollack AS, Vennam S, Gong X, West RB, Pollack JR. New ameloblastoma cell lines enable preclinical study of targeted therapies. Journal of Dental Research. 2022 Nov;101(12):1517-25.