World Journal of Advanced Research and Reviews
eISSN: 2581-9615 CODEN (USA): WJARAI
Cross Ref DOI: 10.30574/wjarr
Journal homepage: https://wjarr.com/

(CASE REPORT)

A rare case of ameloblastoma arising from radicular cyst in mandible

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World Journal of Advanced Research and Reviews, 2025, 25(01), 1322-1328

Publication history: Received on 01 December 2024; revised on 13 January 2025; accepted on 15 January 2025

Article DOI: https://doi.org/10.30574/wjarr.2025.25.1.0095

Abstract

Background: A benign odontogenic tumour that frequently affects the jaw is ameloblastoma. There are only a small number of variations whose biological profile is not elicited, aside from the most often observed clinicopathologic models. When a cystic lesion exhibits the clinical, radiological, or physical characteristics of a mandibular cyst, it is referred to as a unicystic ameloblastoma. However, a histologic investigation reveals a typical ameloblastomatous epithelium lining the cyst cavity, with or without luminal and or mural tumour development.

Purpose: The purpose of writing this article is to describe a rare case of ameloblastoma arising from radicular cyst in mandible.

Case: A 33 year old man presented a lump in his right posterior mandible. Panoramic radiography and CT Scan image revealed a mass involving his right mandibular. The results of an incisional biopsy lead to radicular cyst. Decompression of cyst was performed, after 2 years, there was a slight shrinkage of the tumor and the results second biopsy showed an ameloblastoma.

Case management: En-bloc resection and reconstruction were performed on the patient. Following the final procedure, healing went smoothly, and the patient is currently two years into a postoperative phase during which there have been no recurrences.

Conclusion: The most frequent epithelial odontogenic tumour is the ameloblastoma, however it only accounts for 1% of all jaw tumours and cysts. It is still debatable whether or not the ameloblastoma develops from the radicular cyst. Therefore, the most accurate method for distinguishing unicystic ameloblastoma from radicular cysts is a histological study

Keywords: Radicular cyst; Ameloblastoma; En-block mandibular resection; Medicine

1. Introduction

Ameloblastoma is the most widely recognized clinically significant odontogenic tumour. It addresses around 1% of oral ectodermal growths. Ameloblastoma are exclusively found in the mandible or lower jaw, near the angle area. Presurgical making arrangements for maxillofacial medical procedure involves clinical assessment, investigation of various conventional radiographs, and computed tomography (CT) and magnetic resonance imaging (MRI). Treatment for

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ameloblastoma shifts from curettage to en bloc resection; remodelling of the impacted region is essential after en alliance resection [1,2].

Ameloblastoma type denotes many clinicopathological and histological subtypes. It can be clinically categorized into solid, unicystic, peripheral, and desmoplastic forms and histologically classified into granular, follicular, basal cell, acanthomatous, and desmoplastic [3]. A granular cell pattern in ameloblastoma represents a distinct histological variant of unicystic/multicystic ameloblastoma. It is uncommon and constitutes approximately 3–5% of occurrences. This granular cell ameloblastoma resembles others clinically or radiographically, but its distinctive characteristics may only be discerned through histopathological examination [1,4].

Histopatological traits include granular alterations in stellate reticulum-like cells found within epithelial follicles. Granular cell clusters are full of cytoplasm and include eosinophilic granules. Granular cells are seen in the centre mass of epithelial tumour islands and cords. The island's margin is made up of columnar non-granular cells. According to research, granularity is caused by cytoplasmic excess. Long-standing lesions may also be caused by ageing or degenerative processes. Granular cell ameloblastoma has an aggressive habit with a significant likelihood of recurrence, particularly following suboptimal surgical therapy, and may progress to metastases [1].

While infrequent, certain instances present an elevated risk of a radicular cyst evolving into ameloblastoma; likewise, the inflammatory process has been associated with the pathogenesis of ameloblastoma, especially the unicystic variant that develops from the neoplastic transformation of the non-neoplastic epithelial lining of odontogenic cysts, including radicular cysts, dentigerous cysts, and odontogenic keratocyst. Consequently, postoperative follow-up is critically important [4,5].

2. Case Report

A male patient with an age of 33 years came to Unair Hospital with a lump in the right posterior jaw region which was slowly growing in size since 2018. None of the past medical, surgical, personal or family history was contributing. The patient had a history of 2 biopsy operations at 2 different places. The first PA review was performed with the results of radicular cysts in June and July 2019. Then an incisional biopsy was performed again and the results were unicystic ameloblastoma. The patient has a history of obturator insertion and cyst reduction since August 2019. His history of high blood pressure, diabetes, asthma, and drug allergies was denied.

On physical examination, the patient was in good general condition, the general status was within normal limits. The results of the extraoral clinical examination (figure 1) show an asymmetrical face with a palpable mass in the right mandibular region, no ulcers or fistulas, and the colour is the same as the surrounding tissue. Palpation indicated a palpable mass in the right mandibular region, no palpable oedema or pain, a palpable inferior border of the jaw, no paresthesias in the right mandible, and no palpable enlargement of regional lymph nodes.

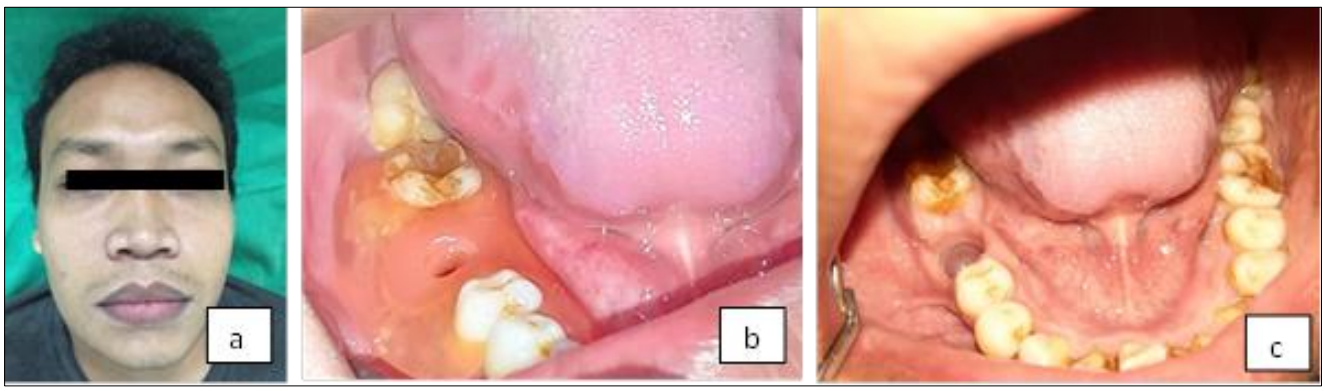

Figure 1 a. Front view; b. Intra-oral tooth region 46 (with obturator); c. Intra-oral tooth region 46 (without obturator)

On intraoral examination (figure 1b 1c): The marsupial canal is visible in region 46 with a mucosal base, no fluid production is seen, the color is the same as the surrounding mucosa, there is no lingual or buccal expansion of the mass, no ulcer is seen, no caries or recession is seen. gingiva on teeth 45, 47, 48. Caries was seen on the occlusal teeth of 17, 26, 36. Palpation examination palpable marsupial canal in region 46 with a mucosal base, palpable buccal crepitus 48,

no palpable lingual mass expansion, no palpable tenderness, no mobility of teeth 45, 47, 48, vitality of teeth Chlor ethyl 45, 47, 48 (+) probe teeth 17, 26, 36 as deep as the pulp, no pain on the probe and druk 17, 26,36.

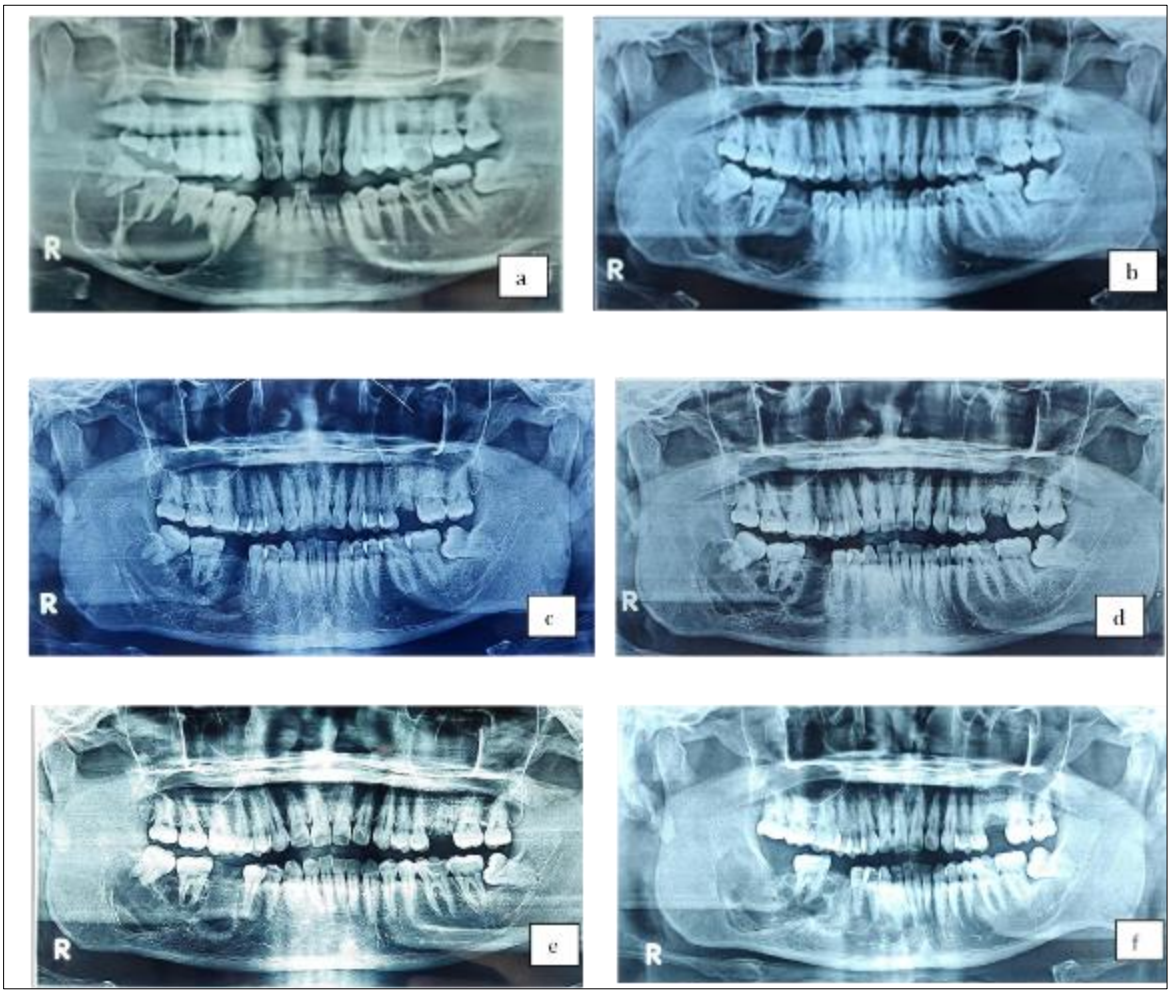

Figure 2 Panoramic Radiology Photo Series. a. June 2019 ; b. December 2019 ; c. August 2020 ; d. November 2020 ; e. February 2021 ; f. June 2021/ OPG of the first visit

Panoramic photos of the patient were taken serially from the time he came and he felt a mass in the right mandible in June 2019. There was a radiolucent image in the region of the lower right second premolar to the periapical area of the lower right third molar pressing against the roots of the primary teeth (Fig. 2a). Then an evaluation was carried out after the extraction of tooth 46, incisional biopsy and marsupialization by placing an acrylic plate 6 months later in December 2019, the impression of a reduced mass was obtained (figure 2b). Evaluation in August 2020, it was found that the impression of the mass was shrinking and there was a process of reinforcement (figure 2c). Then 3 months of evaluation, multilocular lesions were seen at the apex of teeth 47, 48 (figure 2d) and an evaluation 3 months later in February 2021 showed the same impression on the previous panoramic view (figure 2e). Then a post-reinstitutional biopsy was evaluated and tooth extraction 48 in June 2021 showed a multilocular lesion in the apex region of 44 extending to the right angle of the mandible, clear boundaries, no root resorption, and intact mandibular border bone (figure 2f).

On the CT-Scan image of the head without contrast in July 2019 there was a mass in the angle to the right mandibular body with expansion causing cortical thinning, no septation or fluid level was seen, gas forming and correlation with the root of the tooth with the impression of a dentigerous cyst in the angle to the corpus right mandible (figure 3). Then an evaluation was carried out after (figure 4) post extraction of tooth 46 and marsupialization by installing an acrylic plate in March 2021, it was found a cystic mass (7-16 HU), expansive, partially undefined, there was septated in the angle to the right mandibular body causing cortical thinning, and found a correlation with the root of the tooth which showed a dentigerous cyst at the angle to the right mandibular body (when compared to head CT - 05 July 2019 - same impression).

Figure 3 Head CT Scan Evaluation on 5th July 2019

Figure 4 Head CT Scan Evaluation on 1st March 2021

On examination, the biopsy was performed three times. First, the cyst wall tissue was partially lined by odontogenic epithelial cells, fibrous connective tissue with lymphocytic inflammatory cell infiltration and cartilage tissue appeared. It can be concluded that a benign odontogenic cyst can be found in the radicular cyst. Then a biopsy was performed again which showed that the granulation tissue was chronically inflamed, it could be part of a radicular cyst, which was concluded to be a benign odontogenic cyst, which could be found in a radicular cyst. One year later, another biopsy was performed. The cyst wall was covered with odontogenic epithelial cells with a round nucleus, fine chromatin, sufficient cytoplasm, stroma with myxoid areas and inflammatory cell infiltration of lymphocytes and histiocytes, it can be concluded that ameloblastoma is unicystic type.

3. Case Management

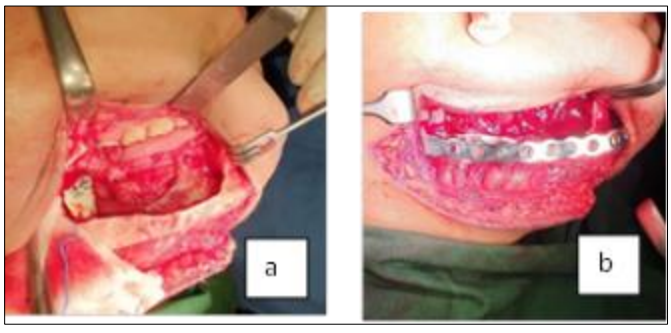

Figure 5 a. Defect post op en-bloc right mandibular resection ; b. Stabilization plate installation

The patient was treated surgically with en-bloc resection of the right mandible with the anterior resection border of tooth 44 to the right ascending ramus of the mandible, stabilization with a reconstruction plate. Histopathological analysis of the tumor mass was carried out and obtained a picture of tissue with tumor growth arranged in anastomosing bands and partly in islands, consisting of a proliferation of odontogenic cells with round oval nuclei, fine

chromatin, sufficient cytoplasm, palisading margins, The middle part appears loose forming a picture resembling the stallate reticulum and it is concluded that ameloblastoma is plexiform and follicular type. Surgical specimens obtained tissue weighing 12 grams with size 0.5x0.5x3.5x2.5x0.5 cm, soft solid consistency partially hard solid, white gray color. From the results of the evaluation of panoramic radiographs on the 7th postoperative day, it was found that the postoperative defect was from the mesial marginal region of the tooth 42 to the posterior 2/3 of the mandibular body, the reconstruction plate, screws, and grafts were well installed, the archbar pond on the left RA and RB teeth. with intermaxillary fixation.

Figure 6 Panoramic X-ray evaluation postoperative h+7

Before surgery, the surgeon used the initial biomodel with the tumor to mark the areas where osteotomies should be performed simulated which the safety margin determined. The tumor was resected and submitted to his-topathological examination. The model was used during surgery as a guide for the surgeon to reconstruction with plate recontrustion so *more precise results and no recurrences.*

4. Discussion

Ameloblastoma is categorised as follows according to the 2005 WHO classification based on differences in biologic behaviour, treatment strategy, and recurrence rate: Traditional solid/multicystic, unicystic, peripheral, and desmoplastic lesions, including so-called hybrid lesions and cancerous kinds. According to certain reports, the odontogenic cysts' epithelium can develop into benign odontogenic tumours like ameloblastoma [6].

The causes of ameloblastoma resulting from odontogenic cysts have been theorised to include nonspecific irritant factors (such as extraction, trauma, infection, inflammation, and unerupted teeth), nutritional deficiency problems, and viral infection [6]. Ameloblastoma and calcifying odontogenic cyst microscopic characteristics have been shown in numerous reports in the literature to merge into one another. Small islands of ameloblastomatous epithelium in unicystic ameloblastomas are sometimes mistaken for radicular cysts or keratocysts, as in our instance [7].

The pathogenesis of cystic ameloblastomas is unclear. Some experts believe they are caused by pre-existing odontogenic cysts, while others believe they grow spontaneously. A comparison of the histological characteristics of the cystic lining of a radicular cyst with those of a cyst exhibiting ameloblastomatous change revealed a focal transformation of stratified squamous epithelium to ameloblastomatous epithelium, featuring loosely attached cells resembling stellate reticulum, palisaded cuboidal basal cells demonstrating reverse nuclear polarity, hyperchromatic nuclei, and pale cytoplasm. Chronic inflammatory cell infiltration was observed in the outer fibrous capsule [5].

The initial endeavors focused on the expression of carbohydrates in blood cells. Although it was deemed beneficial at first, it was subsequently refuted. Imaging modalities such as contrast-enhanced MRI have also been recorded. Thick enhancement in the walls of unicystic ameloblastoma has been identified as beneficial. Histoenzymological investigations were conducted to ascertain the alterations in the activity levels of oxidative enzymes, diaphoresis, acid phosphatases, and naphthol esters. The odontogenic cyst exhibited nonspecific activity, while the ameloblastoma demonstrated consistently low oxidative enzymatic activities in the epithelium and extensive alkaline phosphatase activity in the stroma [8].

A few immunohistochemistry markers have also been examined to better understand the pathophysiology of this tumour. The expression of Bcl-2, CD-68, and β-catenin contributes to pathogenic pathways. The immunohistochemistry results in studies highlight Bcl-2 negativity in granular cells suggesting death, CD-68 positivity in granular cells indicating the existence of lysosome aggregates, and β-catenin positive for cytoplasm in granular cells indicating an altered cell signalling pathway. Mouse double minute 2 (MDM2) protein levels were measured in cases of radicular cysts with ameloblastomatous-like changes, as well as cystic ameloblastoma originating from radicular cysts [1].

The therapy of unicystic ameloblastoma is based on pathological and anatomical grounds, with curettage being indicated. When developing a surgical treatment plan, it is important to consider overall health, tumor size, location, length, psychological impact, and frequent follow-up exams [9]. Extensive unicystic lesions should be biopsied at multiple sites to avoid sampling nondiagnostic regions. Treatment might be radical or conservative. Although the radical technique has been shown to have low recurrence rates, it typically leads in morbidity and substantial consequences, even after successful reconstruction. As a result, therapy should be as conservative as possible, which is backed by the fact that ameloblastoma, particularly the unicystic kind, is a slow growing, locally invasive, and relatively rare metastasizing benign tumor [10].

Case of ameloblastoma by Demir and Gunhan (2023) with conservative approach using marsupialization obturator showed a good healing. The lesion showed a favorable response and shrunk in size following marsupialization. An acrylic obturator was designed to maintain the lesion uncovered and prevent food impaction. Prior to the application of the acrylic obturator, the marsupialization orifice was maintained with daily replaced gauze coated in topical antibiotic ointment. Following the placement of the acrylic obturator, the patient was urged to uphold comprehensive dental hygiene by self-irrigating the cavity daily [11].

Radical and aggressive surgical intervention is the primary approach for managing recurrent ameloblastoma. This approach advocates that mandibular resection must extend at least 1–2 cm beyond the radiological boundary to guarantee the complete excision of all microlesions. In multiple instances, a radical therapeutic approach involving segmental mandibulectomy was chosen due to aggressive characteristics, including cortical bone puncture, tumor expansion, dental nerve infiltration, and multicystic morphology. In all other instances, a marginal mandibulectomy was chosen, considering the preservation of adjacent anatomical components, including the oral nerve and basal mandibular cortex[12].

Irrespective of the treatment method implemented, a rigorous follow-up protocol consisting of monthly assessments during the initial year post-surgery, followed by evaluations every three months, six months, or year thereafter, must be meticulously observed [10]. The distinction between ameloblastomas with cystic degeneration, those originting from odontogenic cysts, and those with ameloblastomatous hyperplasia in odontogenic cysts is now a source of debate regarding this tumour.

5. Conclusion

The most frequent epithelial odontogenic tumour is the ameloblastoma, however it only accounts for 1% of all jaw tumours and cysts. It is still debatable whether or not the ameloblastoma develops from the radicular cyst. Therefore, the most accurate method for distinguishing unicystic ameloblastoma from radicular cysts is a histological study.

Compliance with ethical standards

Disclosure of conflict of interest

There is no conflict of interest.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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