CASE REPORT

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Recurrent follicular ameloblastoma of the maxilla: a case report

Hasmawati Hajar[1*], Barunawaty Yunus[2], Mohammad Gazali[3]

ABSTRACT

Objectives: This report aims to describe the clinical and radiological characteristics of ameloblastoma. This benign odontogenic tumor rarely occurs in the upper jaw, and to emphasize the importance of using panoramic radiography in confirming the diagnosis.

Case Report: A 52-year-old female patient presented with a lump in the right upper jaw that had been present for three months. She reported that a small lump first appeared four years earlier and had gradually increased in size. The patient had a history of surgery to remove a lump and extraction of a right maxillary tooth in 2021, with histopathological analysis (HPA) revealing ameloblastoma. She also reported tenderness in the area and the presence of a salty discharge from the mouth. Panoramic radiography and CT scan revealed a unilocular, homogeneous radiolucent lesion, ovoid with well-defined margins, located in the edentulous posterior right alveolar ridge. The lesion extended superiorly toward the base of the right maxillary sinus and inferiorly toward the crest of the alveolar ridge. The patient underwent an excisional biopsy, with a histopathological examination confirming the diagnosis of a bone tumor (ameloblastoma). This was followed by a segmental maxillectomy in the region of teeth 12–15 under general anesthesia. Ameloblastoma has a high tendency to recur, particularly when conservative management fails to completely remove the tumor. In this case, pathological examination, along with panoramic and CT imaging, confirmed recurrent ameloblastoma.

Conclusion: The diagnosis of recurrent follicular ameloblastoma of the maxilla was established through a combination of clinical evaluation, radiographic imaging, and histopathological examination.

Keywords: *Ameloblastoma, follicular, maxillary, panoramic radiography*
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INTRODUCTION

One of the most common tumors of the jaw is ameloblastoma. This tumor was first recognized by Cusack in 1827 and later described by Broca in 1868.[1] Ameloblastoma accounts for approximately 30% of all benign tumors originating from the odontogenic region of the jaw.[2] It is a benign odontogenic tumor that is locally aggressive and originates from the epithelium.[3] Ameloblastoma is a slowly growing, locally invasive tumor that affects the maxillofacial region. There are various histological variants of ameloblastoma, the most common being follicular ameloblastoma. This variant typically occurs in older individuals and has a high recurrence rate.[4,5]

Based on its histological type, ameloblastoma can be classified into several variants, including follicular, plexiform, acanthomatous, basal cell pattern, unicystic, granular cell pattern, papilliferous ameloblastoma, hemangioameloblastoma, desmoplastic, plexiform unicystic, benign cell, dentinoameloblastoma, melanoameloblastoma, and keratoameloblastoma.[5-7] The most common histological variants are the follicular and plexiform types. Follicular ameloblastoma, also known as the "classic" or "conventional" type, accounts for approximately 64.9% of cases, followed by the plexiform type (13.0%), desmoplastic (5.2%), and acanthomatous (3.9%) variants. Although benign, ameloblastoma can be locally invasive and has a high tendency to recur. Most diagnosed cases occur between the third and fifth decades of life and are rare in children.[8,9]

As ameloblastoma grows to a significant size, it can threaten the airway and digestive system, even increasing the risk of mortality, thereby causing symptoms. Particularly in the upper jaw, due to its coral-like structure, ameloblastoma can reach considerable sizes and spread through the sinuses, orbit, nasal cavity, or skull.[10] According to the latest classification by the World Health Organization (WHO) in 2017, ameloblastoma is divided into four

[1]Oral and Maxillofacial Radiology Residency Program, Faculty of Dentistry, Hasanuddin University, Makassar, Indonesia 90245

[2]Department of Oral and Maxillofacial Radiology, Faculty of Dentistry, Hasanuddin University, Makassar, Indonesia 90245

[3]Department of Oral and Maxillofacial Radiology, Faculty of Dentistry, Hasanuddin University, Makassar, Indonesia 90245

*Correspondence to:
Hasmawati Hajar
hasmawatihajar8@gmail.com

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subtypes: conventional ameloblastoma, unicystic ameloblastoma, peripheral (extraosseous) ameloblastoma, and metastasizing ameloblastoma.[11]

CASE REPORT

A 52-year-old female patient presented to the dental and oral hospital with a complaint of a lump on the upper right side of her mouth that had been present for approximately three months. About four years earlier, the lump had begun as a small, kernel-sized bump and had gradually grown to the size of a meatball. In 2021, the patient underwent surgery to remove the lump, along with the extraction of an upper right jaw tooth. Histopathological examination confirmed the diagnosis of ameloblastoma. The patient reported pain when pressure was applied to the lump area. Occasionally, she experienced a salty discharge from her mouth and felt a sense of increased heaviness on the right side of her face.

On extraoral examination, the face appeared symmetrical. Intraoral examination revealed an enlargement of the right maxillary arch in the region of teeth 12–15, measuring approximately 3 × 2.3 × 2 cm, with a hard consistency. No crepitation was detected, but erythema and tenderness on palpation were present. The affected tissue appeared redder than the surrounding tissue. Calculus deposits were observed, and oral hygiene was poor (Figure 1).

The patient underwent a panoramic examination, which revealed a radiolucent, homogeneous, unilocular lesion with well-defined borders extending from the posterior right alveolar ridge superiorly, nearly reaching the floor of the right maxillary sinus. There was evidence of the destruction of the posterior alveolar ridge extending toward the right maxillary tuberosity.

A CT scan showed a hypodense lesion with sharp borders, smooth edges, and an ovoid shape, measuring approximately 1.4 × 1.6 × 1.5 cm within the right maxillary bone. No pathological hypodense or hyperdense lesions were observed intracranially. The suspected radiodiagnosis was a benign bone tumor in the right maxillary region involving teeth 12–15, with a differential diagnosis of ameloblastoma. The patient underwent surgical excision followed by biopsy. Histopathological analysis confirmed a diagnosis of follicular ameloblastoma. The next treatment plan was a partial maxillectomy involving the region of teeth 12–15 on the right side.

DISCUSSION

Ameloblastoma is a persistent and locally invasive tumor. The most common etiological factors include trauma, inflammation, chronic

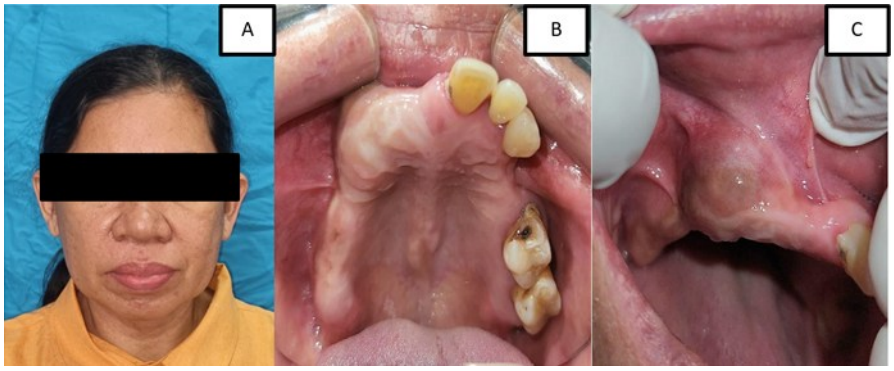

Figure 1. A. Clinical extraoral examination (face appeared symmetrical), B and C. Clinical intraoral examination (enlargement of the right maxillary arch in the region of teeth 12-15 with a hard consistency)

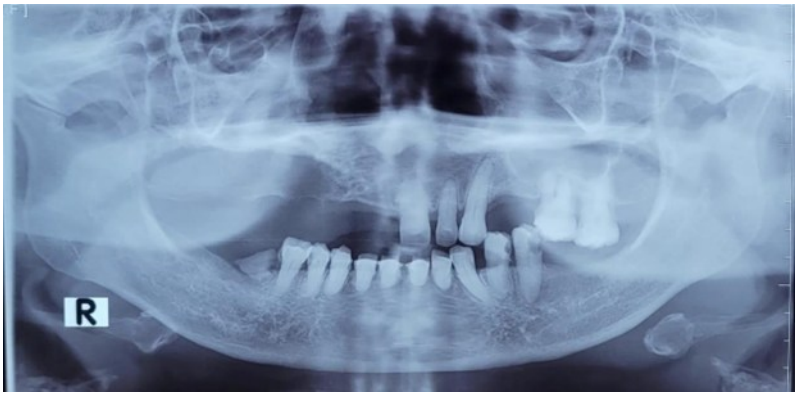

Figure 2. Panoramic View: A radiolucent homogeneous unilocular lesion with well-defined borders extending from the posterior right alveolar ridge towards the superior direction, almost reaching the floor of the right maxillary sinus

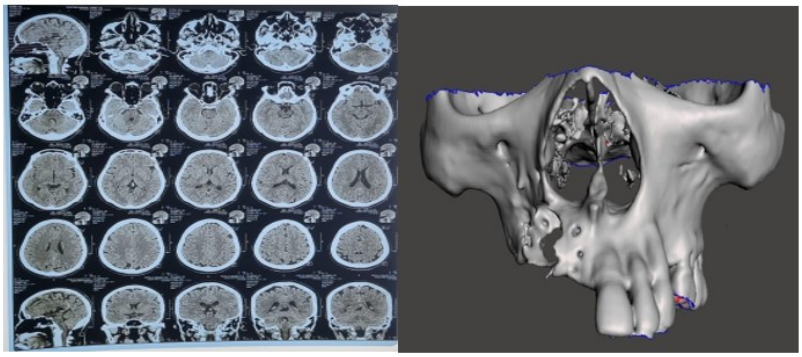

Figure 3. CT Scan Axial View Showed a hypodense lesion with sharp borders, smooth edges, ovoid shape, measuring approximately 1.4 x 1.6 x 1.5 cm in the right maxillary bone

irritation, and infection, often with a history of traumatic injury or tooth extraction. [4] Most cases are asymptomatic and are detected during routine dental X-rays. Swelling or slow-growing jaw expansion without pain, resulting in facial asymmetry, is a common symptom. Dental malocclusion may also occur. The lesion can erode the bone and extend into the surrounding soft tissues.[12] In their analysis of recurrent ameloblastoma, Vydia et al. found that a multilocular radiographic appearance, follicular histopathology, and conservative treatment are risk factors for recurrence. Conservative management was associated with a significantly increased recurrence rate. Although the majority (62%) of analyzed cases were unilocular ameloblastomas, multilocular lesions showed a higher recurrence rate.[10]

The recurrence rate of ameloblastoma is approximately 13%–15% after surgical resection, whereas it ranges from 90%–100% after curettage. The recurrence rate is higher in mandibular ameloblastomas and in the follicular histological type, compared to maxillary ameloblastomas and other variants such as the plexiform type. Unicystic ameloblastoma, characterized by a fibrous capsule, has a significantly lower recurrence rate. Solid or multicystic ameloblastomas tend to be locally invasive, with a recurrence rate of up to 90% following conservative treatments such as curettage and enucleation.[12] Ameloblastoma is a rare odontogenic epithelial neoplasm, accounting for only 1% of all upper jaw tumors and 11% of odontogenic tumors.13,14 According to published studies, the average age of patients with classic ameloblastoma is generally between 40 and 50 years, with the average age at diagnosis being 40 years. In this case report, the patient is 52 years old. Ameloblastoma of the upper jaw is less common than those occurring in the mandible. Unlike the compact bone of the mandible, the upper jawbone is spongier, allowing the tumor to more easily invade and spread to adjacent structures such as the nasal cavity, paranasal sinuses, orbit, pharyngeal tissues, and skull base. In such cases, ameloblastoma can present various symptoms, including unilateral facial deformity, toothache, headache, nasal obstruction, epistaxis, and visual disturbances. As seen in this patient, pain or tenderness may occur upon the palpation of the affected area. For descriptive purposes, maxillary ameloblastomas are classified as either anterior or posterior tumors, with the posterior region being more commonly affected.[14,15]

Radical surgery is the current standard treatment for ameloblastoma, depending on the histological subtype and the surgical technique used to achieve clear margins free of neoplastic cells, thereby reducing the risk of local recurrence.[16] The preferred treatment approach is wide local excision with clear margins, followed by immediate reconstruction. Partial maxillectomy, performed with a safety margin of 10–15 mm on healthy bone, including the alveolar ridge, hard palate, maxillary sinus mucosa, and lateral nasal wall, is commonly employed. In this case, the treatment plan is partial maxillectomy. To minimize the risk of recurrence, it is essential to identify the clinical factors associated with ameloblastoma recurrence. Rong et al. reported that recurrence is linked to several factors, including tumor location, root resorption, and invasion of the maxillary sinus, regardless of the surgical method used.[14,17]

In general, ameloblastomas on radiographic examination typically present as radiolucent cystic lesions, either unilocular or multilocular, with features resembling a "soap bubble" appearance. Additional characteristics include cortical thinning or destruction, local invasion, and root resorption. In this case, the lesion is classified as a unicystic ameloblastoma, with histological examination revealing the follicular subtype. Radiographic findings include a homogeneous radiolucent unilocular lesion with well-defined borders, extending from the posterior right alveolar ridge superiorly, nearly reaching the floor of the right maxillary sinus, and accompanied by cortical destruction. CT scans are the most reliable modality for identifying cortical damage and soft tissue involvement due to tumor cell infiltration, particularly into the cancellous portion of the cortical bone.[12,13] Benign tumors generally grow slowly by forming additional internal tissue, resulting in borders that appear relatively smooth, well-defined, and sometimes corticated. The internal structure of such tumors may be radiolucent, radiopaque, or a combination of both. In this case, imaging shows cortical destruction associated with a hypodense lesion characterized by sharp borders, smooth edges, and an oval shape.

Among all histopathological variants of ameloblastoma, the follicular pattern is the most common. The most frequent differential diagnoses for ameloblastoma in the posterior mandible include odontogenic keratocyst, central giant cell granuloma (CGCG), odontogenic myxoma, primary and secondary hyperparathyroidism, central hemangioma, and aneurysmal bone cyst.[17,18]

CONCLUSION

Ameloblastoma is the most common odontogenic tumor, typically affecting individuals in advanced age. It can occur in both the upper and lower jaws, but it more frequently presents in the lower jaw. There are numerous variants of Ameloblastoma distinguished by histological patterns, with the follicular variant being the most prevalent. Prompt management of Ameloblastoma is essential upon detection, as it can lead to facial deformities in the long term. The management approach depends on the histological variant and the involved anatomical structures to prevent recurrence. Long-term follow-up required in ameloblastoma to check for recurrence.

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FOOTNOTES

All authors have no potential conflict of interest to declare for this article. Informed consent was obtained from the patient for being included in this case report.

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