CASE REPORT

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Radiographic imagery of aggressive plexiform-type ameloblastoma in the mandible: a case report

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ABSTRACT

Objectives: This case report aims to describe the radiographic characteristics of plexiform ameloblastoma and its impact on surrounding tissues in a middle-aged female patient using panoramic radiography and computed tomography, along with the case management.

Case Report: A 43-year-old female patient presented to the Dental Radiology Unit of RSGMP UNHAS with a panoramic referral letter, diagnosed clinically with anterior mandibular ameloblastoma. Extraoral examination revealed an asymmetrical facial appearance with anterior mandibular enlargement. Intraoral examination showed mucus membrane enlargement in the anterior mandible region (teeth 37-45), soft consistency, absence of crepitus, no palpation tenderness, and mobility in several anterior mandibular teeth. The first panoramic radiograph (March 16, 2022) exhibited a unilocular radiolucent lesion, well-defined, with scalloped margins in the anteroposterior mandibular region. The second panoramic examination (May 23, 2022) indicated a more aggressive lesion expansion, with evidence of root resorption and destruction of the inferior mandibular cortex approaching the mandibular angle. CBCT findings demonstrated a hypodense/radiolucent lesion extending anteroposteriorly, superiorly, and inferiorly, leading to displacement, root resorption, and destruction of the inferior mandibular cortex in the inferior direction.

Conclusion: Based on the characteristics and structure of the lesion observed through various radiographic examinations, a unilocular ameloblastoma was suspected. Histopathological examination confirmed the plexiform-type ameloblastoma.

Keywords: *Plexiform ameloblastoma, computed tomography, jaw tumor, mandible*
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INTRODUCTION

Ameloblastoma is an aggressive neoplasm arising from remnants of the dental lamina and enamel organ (odontogenic epithelium). It is characterized by aggressive yet benign growth, slow and locally invasive.[1,15] Historically, ameloblastoma has been known for over half a century. The term "ameloblastoma" was suggested by Churchill in 1934.[2] This tumor originates from the development of enamel organ tissue type that doesn't differentiate into enamel formation, as per the WHO definition.[3]

Clinical presentation of this lesion is usually asymptomatic and can be discovered during routine radiographic examination or due to asymptomatic jaw expansion. Tooth movement or malocclusion can sometimes be early signs. Typically, slow growth, painless swelling causing facial asymmetry, large lesions can lead to lose teeth, pain, paresthesia, and ulceration or nasal region obstruction.[2,3]

Radiographically, ameloblastoma is commonly found in bone and detected through dental X-rays or panoramic (orthopantomogram/OPG) images specifically.[5] Radiographically, ameloblastoma appears osteolytic and is typically located in the tooth-bearing areas. Due to its slow growth, margins are usually well-defined and sclerotic.[6] Ameloblastomas often form well-defined radiolucent regions, similar to cysts, with well-defined margins.[7] The slow growth rate is generally associated with root movement. Root resorption can sometimes occur in relation to ameloblastoma growth.[5,6] Solid/multilocular ameloblastomas typically exhibit soap bubble or honeycomb appearances. Computed Tomography (CT) or Cone Beam Computed Tomography (CBCT) can aid in surgical planning by revealing cortical destruction and soft tissue extension. A common appearance is a unilocular or multilocular radiolucent lesion with well-defined borders.[8]

There are six histological subtypes of ameloblastoma, including follicular, plexiform, acanthomatous, granular cell, basal cell, and desmoplastic. Plexiform ameloblastoma is one of the variants with specific histopathological characteristics, featuring a fishnet-like pattern of

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proliferating epithelial cells. This lesion manifests as a unilocular or multilocular radiolucent area in the mandible or maxilla.[4,12]

This article aims to report a rare case of plexiform ameloblastoma in a female patient based on radiographic characteristics from panoramic and CBCT images.

CASE REPORT

A 43-year-old female patient presented to the Dental Radiology Unit of RSGMP UNHAS with a referral letter from an Oral Surgeon for an orthopantomogram examination with a 2-month interval and MDCT in the following month. Anamnesis results from approximately 2 years ago indicated complaints of pain and swelling in the anterior lower jaw. The patient's general condition was good, with no drug allergies and no history of systemic illness. Extraoral examination revealed pain, swelling in the anterior lower jaw, and asymmetrical facial appearance (Figure 1).

Upon intraoral examination, there is enlargement of the anterior mandibular mucosa in the tooth region 37-45 with dimensions of approximately 12.4 x 5.2 x 2.5 cm, soft consistency, no crepitus, color and temperature similar to surrounding tissue, no palpation tenderness, necrotic tissue in the gingival region of teeth 34 and 35, mobility in teeth 31, 32, 37, 42, 43, 41, 44, and 45, along with poor oral hygiene conditions (Figure 2).

The result of the first orthopantomograph examination (March 16, 2022) showed a well-defined, corticated unilocular radiolucent lesion in the anterior mandibular region extending up to (Figure 3a). The following month, the second orthopantomograph examination (May 23, 2022) revealed the progression of a highly aggressive lesion, irregularly unilocular, corticated in all directions anteroposteriorly, superior-inferiorly, and mediolaterally, accompanied by displacement and resorption of the remaining mandibular tooth roots (Figure 3b).

Figure 1. Extraoral view of the patient (a, c) lateral view; (b) frontal view; (d) view from below showing facial asymmetry due to swelling in the mandibular region

Figure 2. Intraoral views of the patient: (a) Front view; (b) Upper jaw; (c) Lower jaw

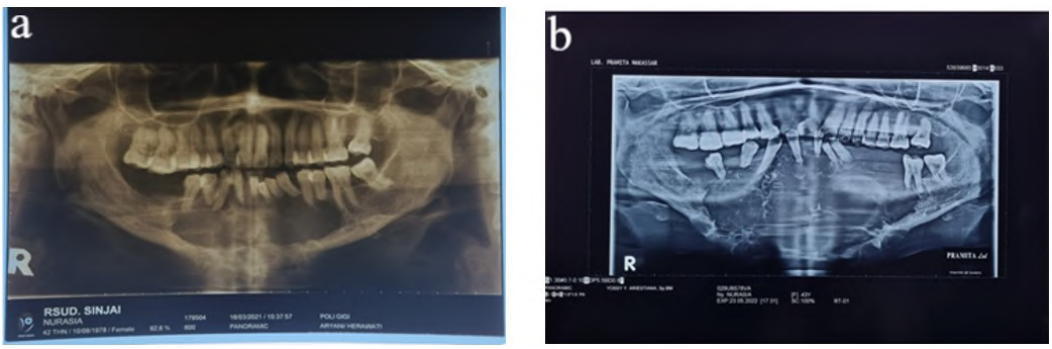

Figure 3. (a) First panoramic radiograph (March 2022) (b) Second panoramic radiograph (May 2022) showing aggressive lesion in the anterior mandibular region within a one-month interval

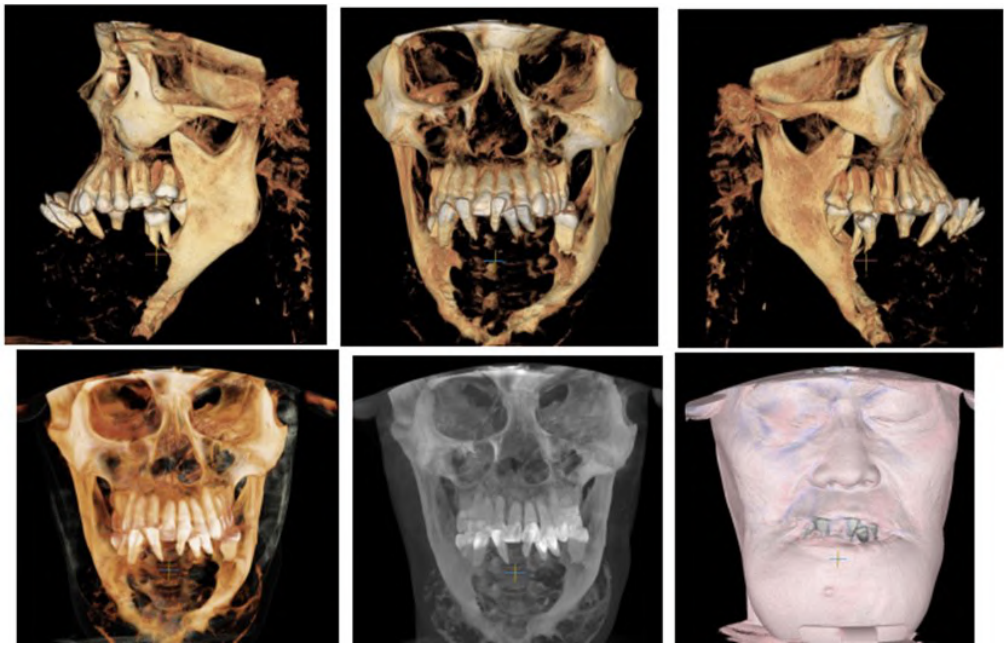

Figure 4. Multiplanar CBCT 3D reconstruction view of bone, bone-soft tissue, and soft tissue

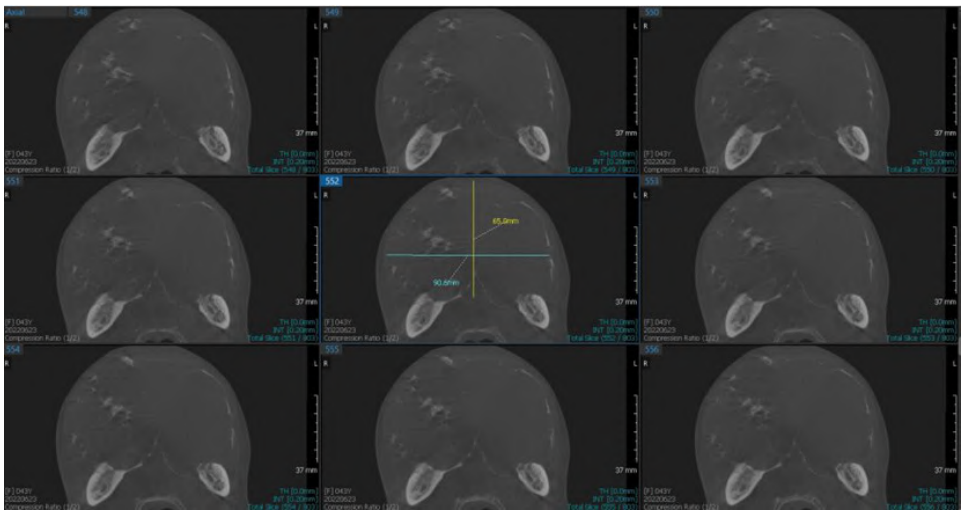

Figure 5. Axial view showing a hypodense lesion measuring approximately 90.6 mm x 65 mm

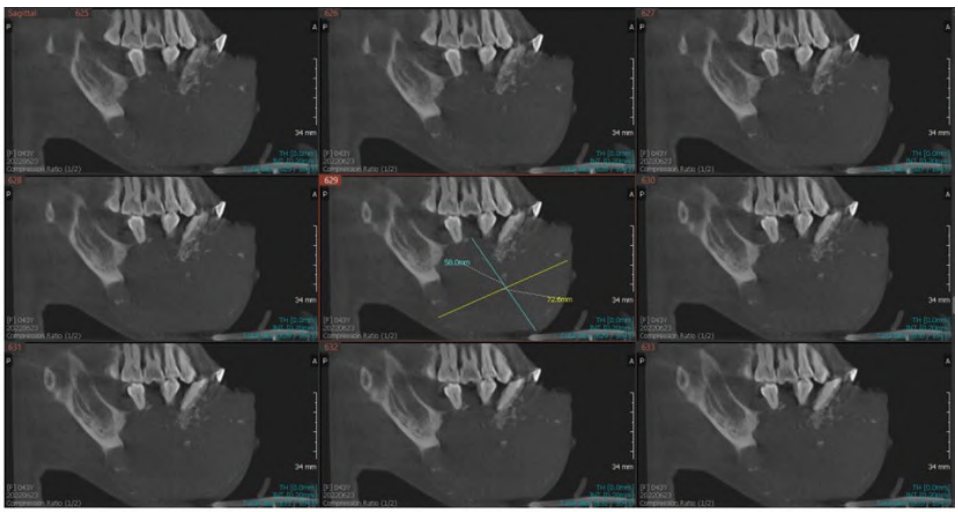

Figure 6. Sagittal view showing lesion dimensions of approximately 72 x 58 mm

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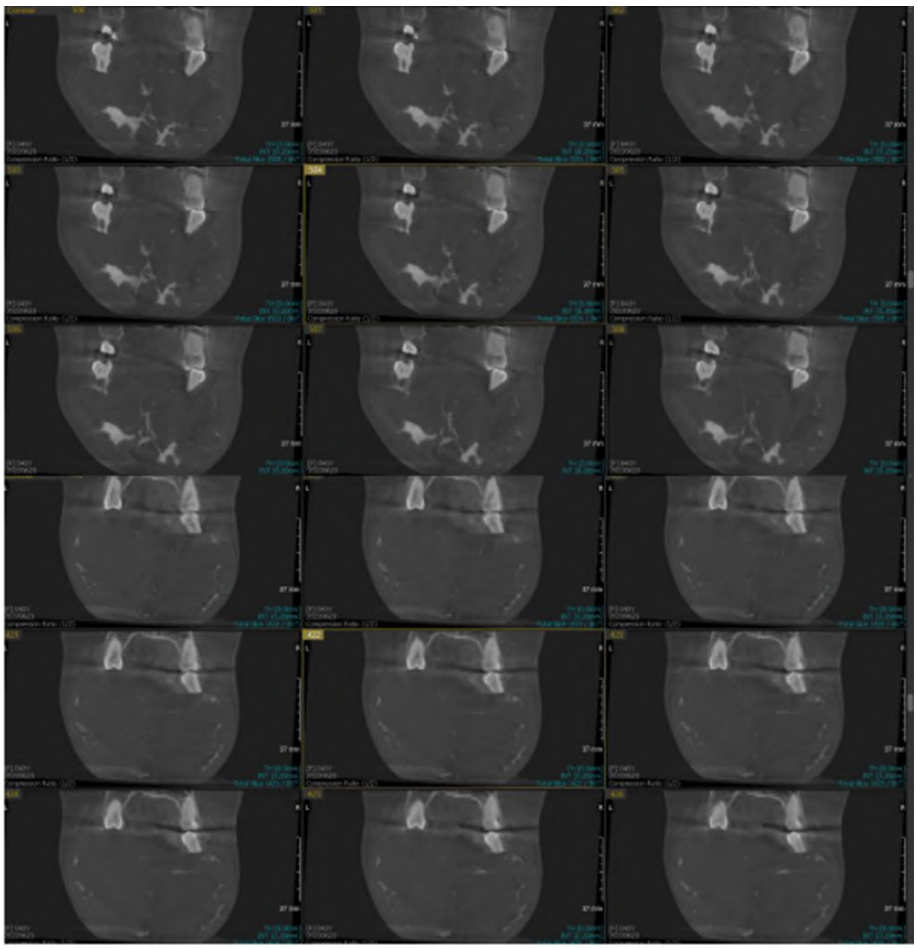

Figure 7. Coronal view of CBCT images

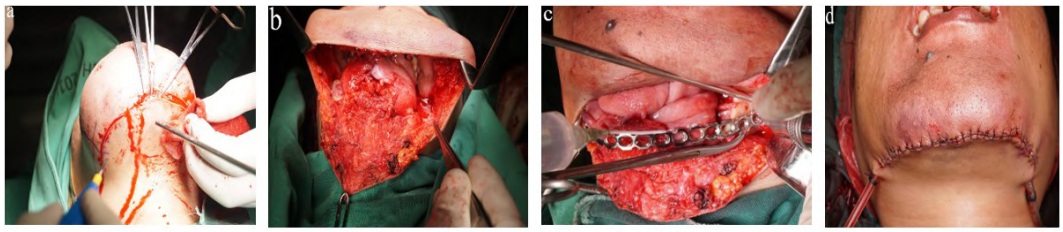

Figure 8. (a) Extraoral Incision; (b) Mandibular Segmental Resection; (c) Insertion of Reconstruction Plate; (d) Suturing

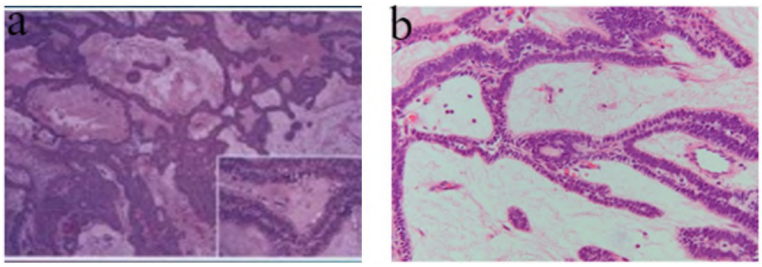

Figure 9. (a) Plexiform ameloblastoma with a proliferation of epithelial cells resembling a fishnet pattern; (b) Plexiform ameloblastoma with string-like strands that anastomose with tumor cells

In the axial view of the mandible, the lesion demonstrates expansion in the anterior-posterior and medio-lateral directions. The bone in the parasymphyseal region and mandibular body appears extensively destroyed (residual bone fragments in the inferior mandibular cortex) with remaining bone in the right-left ramus of the mandible; the lesion measures approximately 90 x 65 mm in the axial view (Figure 5).

In the sagittal view of the right mandible, the lesion extends superiorly-inferiorly from the tooth region to the base of the mandible (tooth 45 is pushed far posteriorly with ½ root resorption, root resorption is also observed in teeth 44-43); the lesion measures approximately 72 x 58 mm in the left sagittal view (Figure 6).

Figure 10. Preoperative intraoral view (Left), Postoperative intraoral view (Right)

In the sagittal view of the left mandible, the lesion extends superiorly-inferiorly from the tooth region to the base of the mandible, with teeth 36 and 37 remaining and root resorption observed on the mesial root of tooth 36); the lesion measures approximately 57 x 55 mm in the left sagittal view (Figure 6).

The MDCT examination results indicated the presence of a primary bone tumor in the mandible. The interpretation of the panoramic radiography and CBCT/MDCT results suggests a suspected radiodiagnosis of this lesion as ameloblastoma. The radiographic findings will serve as diagnostic support for the treatment procedure and will guide the oral surgeon.

The patient underwent a segmental resection surgery under general anesthesia with intubation via tracheostomy. The incision pattern was made approximately 1-2 cm below the mandibular margin, and an extraoral incision was performed using the transmandibular approach. An intraoral incision was made on the right posterior vestibule extending to the vestibule of tooth region 45. Subsequently, a segmental resection of the mandible was performed, followed by the placement of reconstruction plates and suturing (Figure 8). The histopathology examination results reveal the characteristics of plexiform-type ameloblastoma (Figure 9).

DISCUSSION

Plexiform-type ameloblastoma in the anterior mandibular region is a relatively rare occurrence, with only a few reported cases in the literature situated in the anterior region.[4] Steven R Singer et al. explain that plexiform-type ameloblastomas are generally found in the posterior mandible and the remaining cases occur in the maxilla, accounting for 5-15% of cases.[9] Similarly, Vikrant O. Kasat et al. mention that plexiform-type ameloblastomas are commonly found in the posterior mandibular region with an average age of 37.5 years.[10]

Solid-type ameloblastomas can sometimes present with varying radiological appearances in different radiographic examinations, making identification challenging. To establish an accurate diagnosis, it is advisable to employ multiple radiographic techniques.[13] In cases like this, high-resolution modalities such as 3D CBCT radiography are needed to provide a more precise picture. The characteristic radiographic feature of this lesion is its unique location in the anterior mandibular region. Besides its distinctive location, the lesion appears to be destructive and aggressive, leading to mobility and resorption of the remaining mandibular teeth. This differs from the findings of Bina Kashyap et al., where the lesion did not cause tooth mobility or root resorption. Radiographic examinations plays a crucial role in accurately determining the size and extent of the lesion. With the advancement of technology, sophisticated radiographic data such as MDCT and CBCT can guide oral surgeons in treatment planning through 3D reconstruction models, facilitating surgical procedures. Radiographic examination and lesion location are key factors in establishing a diagnosis. Intraoral radiography, panoramic radiography, MDCT/CBCT, and MRI are all used as diagnostic tools. Radiological findings may include expansion of the cortical plate with scalloped margins, multilocularity or a 'soap bubble' appearance, and root resorption.[4] CT is used to depict soft tissue masses, cortical bone damage, and tumor extension into adjacent structures. Although MRI is not commonly used for hard tissue examination, it can provide information about tumor definition and consistency.[11]

The management of plexiform-type ameloblastoma follows the same principles as other types, involving surgical procedures. The goal of ameloblastoma tumor management is complete tumor removal, aesthetic facial reconstruction, a favorable prognosis, and long-term follow-up demonstrating favorable conditions. Solid ameloblastoma typically requires at least jaw resection or excision, as recurrences are reported in 50-90% of cases treated with curettage alone. Resection followed by immediate surgical reconstruction is usually performed for large lesions. Routine follow-up for ameloblastoma cases is essential, as recurrences can be observed up to 10-20 years after primary therapy.[6]

Histological examination is integral in ameloblastoma cases. Many described histological patterns for ameloblastomas may lack clinical relevance. Some present with a single histological type, while others exhibit multiple histological patterns within the same lesion. Generally, all

subtypes show palisading of columnar cells around epithelial nests in a pattern resembling ameloblasts of the enamel organ. The cell nuclei resemble reticulum stellate cells of the enamel organ. Plexiform-type ameloblastomas consist of two to three layers of stellate-like cells at the periphery, forming interconnecting nests.[6,14]

CONCLUSION

Plexiform ameloblastoma is a rare jaw bone tumor with the same etiology as other types of ameloblastoma, typically asymptomatic. The lesion presents as extensive and aggressive on clinical and radiographic examinations, with a specific pattern commonly located in the anterior maxilla or mandible region. Supportive examinations, including radiography and histology, greatly aid in confirming a definitive diagnosis and guiding oral surgeons in managing this lesion.

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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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