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## Solid Multicystic Ameloblastoma Misdiagnosed Radiographically as a Periapical Cyst: A Case Report

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## **CASE REPORT**

# **Solid Multicystic Ameloblastoma Misdiagnosed Radiographically as a Periapical Cyst: A Case Report**

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## **ABSTRACT**

Ameloblastoma is a rare odontogenic neoplasm occurring in the jaw that can be observed as several different histologic and radiographic presentations. Since inadequate treatment is associated with high recurrence rate, accurate diagnosis is critical for proper management of ameloblastoma. The preferred treatment is wide local excision and reconstruction. We herein present a patient with solid multicystic ameloblastoma involving the mandible, which was initially misdiagnosed and treated as a periapical cyst, and discuss various radiographic presentations of ameloblastoma.

**Keywords:** ameloblastoma; multicystic

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## **INTRODUCTION**

Ameloblastoma is a benign odontogenic tumor of epithelial origin that displays a locally aggressive behavior with a high level of recurrence. Ameloblastoma, which shows greater propensity toward the lower molar-ramus region, was first described by Cusack in 1827 and definitively termed as ameloblastoma in 1930 by Ivey and Churchill. Clinically, ameloblastoma can present as unicystic, multicystic, peripheral, or malignant lesions. Radiographically, ameloblastoma appears as a radiolucent lesion with varying sizes and features ranging from a single, well-demarcated lesion to a multilocular lesion with “soap bubble” appearance. We herein describe a patient with a solid multicystic ameloblastoma involving the mandible that was misdiagnosed and treated as a periapical cyst; this case highlights the various radiographic manifestations of ameloblastoma that can aid in its diagnosis.<sup>1,2</sup>

## **CASE REPORT**

A 22-year-old female patient visited our clinic with a chief complaint of painful swelling in the left lower jaw. The swelling, which she noticed two months prior to her visit, was initially small but gradually enlarged

to its current size. In addition, the patient stated the presence of pain for the last five days, which was severe, continuous, and localized, and which was aggravated by chewing on the left side; no relief was achieved with over-the-counter pain medication. Before referral to our clinic, the patient was diagnosed with a periapical cyst by a general dental practitioner, who initiated root canal therapy for these symptoms. The patient’s medical and drug histories were non-contributory.

Extraorally, the swelling was present in the lower third of the left side of the face and measured approximately 3×5 cm, extending from the corner of the mouth to 5 cm ahead of the angle of the mandible. Superiorly, the swelling extended from the line joining the corner of the mouth and the tragus to 2 cm below the inferior border of the mandible. The skin over the swelling appeared normal, and the surface was smooth. On palpation, all inspectory findings were confirmed: there was no local rise in temperature, the overlying skin was pinchable, and the swelling was firm and appeared to be fixed to the underlying structures (Figure 1 a, b).

Intraorally, diffuse swelling, which was tender to touch, was present in the left lower quadrant of the oral cavity which obliterated the buccal and lingual vestibules. The overlying mucosa was erythematous.



**Figure 1.** A. Extraoral swelling on the lower third of the left side of the face; frontal view; B. Extraoral swelling on the lower third of the left side of the face; inferior view; C-D. Intraoral image showing obliteration of the buccal vestibule.



**Figure 2.** Panoramic radiograph obtained before treatment showing periapical radiolucency in teeth #35 and #36. Note the absence of pulpal pathology.

On palpation, there was bicortical expansion in the premolar-molar region, with eggshell crackling in the molar region of the buccal cortex. Grade II mobility was observed in teeth #35 and #36 with tenderness on vertical percussion (Figure 1 c-d). The patient was provisionally diagnosed with ameloblastoma based on the clinical history and findings including the lesion site and the age of the patient.

Panoramic radiograph obtained prior to the initiation of root canal therapy revealed while there were no coronal abnormalities in the affected region, there was a well-defined solitary radiolucency extending from the distal aspect of tooth #34 to the distal aspect of the distal root of tooth #36 periapically. Additionally, root resorption was observed in teeth #35 and #36. The lesion appeared to be uniformly radiolucent with no evidence of septa or calcified specks. Given that the panoramic radiograph was obtained prior to subsequent dental intervention, an intraoral periapical radiograph (IOPAR) as well as occlusal and panoramic radiographs was obtained (Figure 2-5).



**Figure 3.** Intraoral periapical radiograph of teeth #35 and #36



**Figure 4.** Panoramic radiograph showing endodontic intervention.

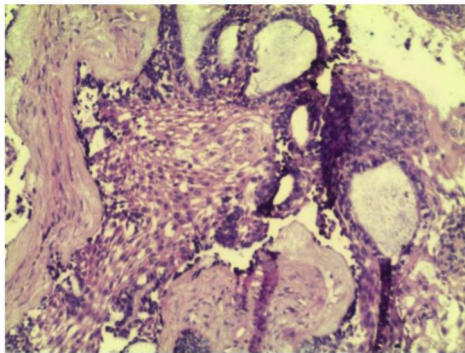


**Figure 5.** Left lateral mandibular cross-sectional occlusal view

IOPAR revealed coronal radiopacity involving the pulp chamber of tooth #36 and radiolucency in tooth #35, indicative of endodontic therapy initiation. Periapically, a well-defined solitary radiolucency was extending from the distal aspect of tooth #34 to the distal aspect of the distal root of the tooth #36. The lesion appeared to be uniformly radiolucent with no evidence of septa or calcified specks. There were no sclerotic borders. The lesion appeared to displace tooth #35 mesially. The lamina dura was lost in teeth #35 and #36, and the roots of teeth #35 and #36 were resorbed. Mandibular lateral occlusal radiograph showed perforation of the buccal cortical plate in the premolar-molar region. In addition to showing findings similar to those observed by IOPAR, panoramic radiograph revealed further that tooth #15 was missing, tooth #53 was retained, and tooth #13 was transposed.



**Figure 6.** Computed tomography showing cortical plate expansion with perforation in the area around teeth #35 and #36



**Figure 7.** Histopathologic examination of hematoxylin and eosin staining of biopsy specimen. Findings are suggestive of solid multicystic ameloblastoma

Contrast-enhanced computed tomography revealed a well-defined multi-loculated cystic lesion containing septations and solid areas, which measured 26×22 mm, in the body of the left mandible. Contrast studies showed enhancement of the wall septations and solid areas within the lesion. No evidence of mineralization was noted. Apex of tooth #36 was observed to be floating inside the lesion. Few lymph nodes measuring 5–7mm in diameter were noted in the submental and submandibular regions. Overall, computed tomography indicated the presence of a multiloculated cystic lesion with enhancing septations as well as solid areas within the body of mandible, suggestive of ameloblastoma (Figure 6).

The specimen obtained by incisional biopsy which was stained with hematoxylin and eosin, revealed the presence of odontogenic epithelium with formation of ducts and solid and sheet pattern. The cells surrounding the lesion were hyperchromatic and appeared tall and columnar in shape. Stellate reticulum-like foci were observed in some areas, whereas other regions contained multicysts. The cystic wall contained connective tissue with dense collagen fibers and exhibited a desmoplastic appearance. Overall



**Figure 8.** Intraoperative images showing removal of the lesion while sparing the lower border of the mandible. An iliac crest graft is used for reconstruction.



**Figure 10.** Panoramic radiograph of the patient at one week after surgery

histopathologic features supported the diagnosis of solid multicystic ameloblastoma (Figure 4).

The lesion was excised in to while retaining the inferior border of the mandible. The mandible was reconstructed using an iliac crest graft, and teeth #35 and #36 were extracted. The patient was followed up for a period of six months during which she remained symptom-free (Figure 7-11).

## DISCUSSION

Multicystic ameloblastoma is a benign epithelial odontogenic tumor of the jaw that is more aggressive than the other ameloblastoma variants, albeit its slow growth, and accounts for approximately 10% of all odontogenic tumors of the jaw.<sup>3-5</sup> The occurrence of ameloblastoma at the apex of teeth is rare and can be often misleading in a definitive diagnosis, as illustrated in this case, which can lead to the initiation of endodontic therapy with the assumption that the lesion is pulpo-periapical in origin, although the pretreatment panoramic radiograph in the current case did not reveal any features suggesting pulp or





Figure 11. Panoramic radiograph at six-month follow-up

periodontal pathology. This initial misdiagnosis was reported previously in similar cases where the definitive diagnosis of ameloblastoma was reached after histological examination. Ameloblastoma can expand, if left untreated, causing the resorption of the roots of associated teeth and tooth displacement, and lead to perforation of the cortical plate which can clinically be elicited by the presence of a sound similar to eggshell crackling, which was present in the current case. Root resorption of the involved teeth, which was present in the current case, is not unique to ameloblastoma. Therefore, it is important for clinicians to be wary of lesions that appear at the apex, which might be of neoplastic origin.<sup>6-8</sup>

Radiographic presentation of ameloblastoma varies, as described by Worth. Unicystic ameloblastoma appears as a unilocular radiolucency resembling a cyst; unlike a cyst, however, discontinuity is observed in the peripheral cortex. Ameloblastoma presenting with a spider web pattern is the most common type, in which the lesion is observed as a large radiolucent area with scalloped borders. From the center of the lumen, coarse strands of trabeculae radiate peripherally, giving it the appearance of a spider web. The soap bubble variant of ameloblastoma consists of multilocular radiolucencies of varying sizes. Ameloblastoma with a honeycomb or solid pattern on radiography appears to have multiple small radiolucencies surrounded by hexagonal bony cortices, giving rise to a honeycomb appearance.<sup>9, 10</sup> Clinicians must be cognizant of the various manifestations of ameloblastoma, with long-term periodic recalls to monitor for potential recurrence after treatment.

## CONCLUSION

Ameloblastoma has a high recurrence rate in patients receiving inadequate treatment. The awareness of various clinical manifestations and radiographic patterns of ameloblastoma should prevent misdiagnosis and incorrect treatment. Ameloblastoma might mimic various pathologies; during radiographic assessment of periapical lesions, however, attention should be paid to the presence of scalloped margins and root resorption.

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