

## CASE REPORT

# An Unusual Case of Ameloblastoma observed in the Left Anterior Mandible

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## ABSTRACT

**Aim:** To report a small intaalveolar ameloblastoma which resembled cystic lesion, and to emphasize the value of Computed tomography (CT) in radiographic examination.

**Background:** Ameloblastoma is a slow-growing, locally invasive odontogenic neoplasm that accounts for approximately 10% of all tumors detected in the jaws. Radiographically, those tumors are usually well-defined. Computerized tomography is highly recommended to confirm the diagnosis.

**Case description:** A 48-year-old female attended with a chief complaint of painless swelling in the left anterior of mandible, 1 month duration. In intraoral examination, non-fluctuant, immobile, approximately 1 × 1 cm in dimension, painless, swelling which had a bone-like hardness and located in the mandibular canine region was detected. Panoramic radiography revealed a well circumscribed unilocular radiolucent lesion located in the inter-radicular area of left mandibular lateral and canine teeth. In CT examination it was realized that the lesion was multilocular. Histopathological examination of the biopsy specimen was reported as ameloblastoma.

**Clinical significance:** It is extremely difficult to find such an ameloblastoma in small dimension in alveolar bone.

**Keywords:** Ameloblastoma, Alveolar bone, Radiographic examination, Computerized tomography.

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## INTRODUCTION

Ameloblastoma, an odontogenic neoplasm, is typically slow-growing, locally invasive and benign. It is the most common clinically significant odontogenic tumor, accounting for approximately 10% of all jaw tumor occurrences. Its exact etiology is unknown.<sup>1-3</sup>

Three clinicoradiographic variants are recognized: solid/multicystic (86%), unicystic (13%) and peripheral (1%). Solid/multicystic ameloblastoma is commonly encountered in patients aged 30 to 70, with neither a gender nor racial predilection. About 85% of all cases are located in the mandible.<sup>3,4</sup>

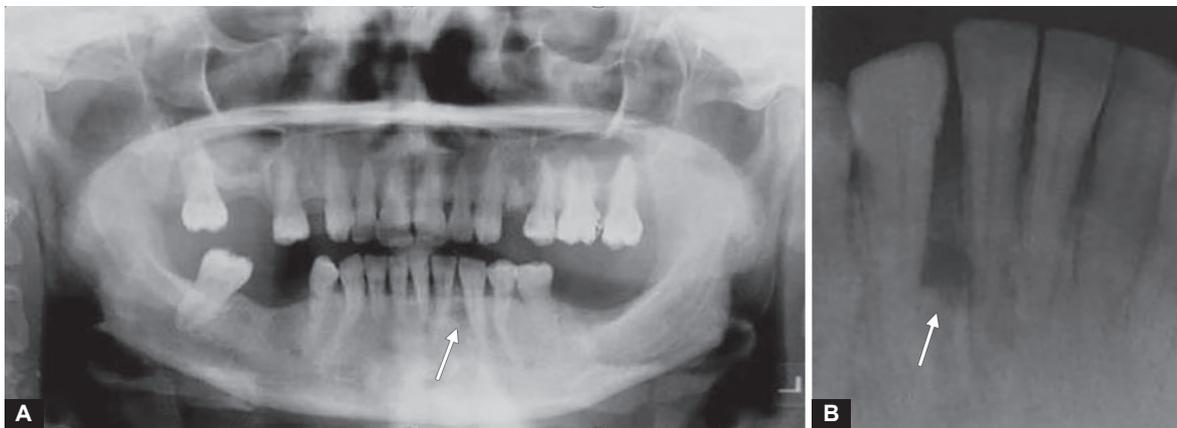
Radiographically, these tumors usually appear well-defined and are frequently delineated by a cortical border. The internal structure varies from completely radiolucent to mixed, with the presence of bony septa that provide multilocular or soap bubble patterns. Occlusal radiography may show bone expansion and thinning of the adjacent cortical plate, and egg shell formation may also be realized. Computerized tomography (CT) is highly recommended if a preliminary diagnosis of ameloblastoma is made, as it not only helps confirm the diagnosis, but also allows for accurate evaluation of the anatomical extent of the tumor and for detecting cortex perforations and soft tissue invasion. CT findings include cystic areas of attenuation with scattered isoattenuating regions, representative of soft tissue components. The lesion can also erode through the cortex, extending into the surrounding oral mucosa. If invasion into soft tissues is extensive, magnetic resonance imaging (MRI) should be performed, as it yields more accurate images of the nature and extent of the invasion.<sup>3,5</sup>

In this report, we aimed to present an extremely rare case of a small ameloblastoma located in the alveolar bone of the anterior mandible, and to confirm the value of CT in the diagnosis of the ameloblastoma.

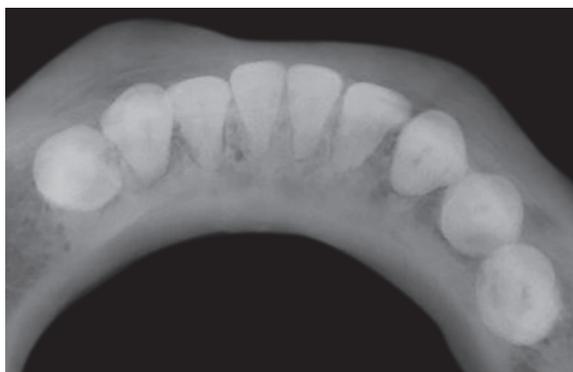
## CASE REPORT

A 48-year-old female presented at the Oral and Maxillofacial Radiology Clinic with a chief complaint of a small painless swelling in the left anterior mandible of 1-month duration.

Her medical history was noncontributory. She had poor oral health, with generalized gingival inflamma-



**Figs 1A and B:** (A) Panoramic radiograph and (B) periapical radiograph showing a well-circumscribed, cyst-like radiolucent lesion, approximately 1 cm in diameter, located between the left mandibular lateral incisor and canine tooth



**Fig. 2:** Occlusal radiograph indicated multiple radiolucencies around the lateral incisor, canine and first premolar teeth but the cortical bone was intact and no bone expansion was noted

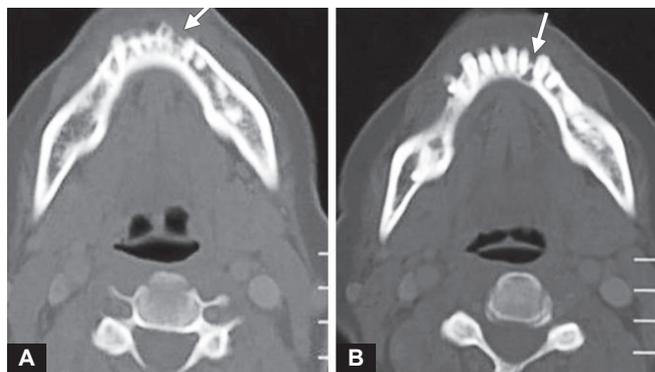
tion and significant tooth loss. Intraoral examination revealed a hard, nonfluctuant and nontender swelling, approximately 1 cm in diameter, located in the mandibular canine tooth region. The relevant teeth were found to be vital.

Panoramic and periapical radiographs revealed a well-circumscribed, unilocular cyst-like radiolucent lesion, located between the left mandibular lateral incisor and the canine. There was no lamina dura loss, displacement or root resorption of adjacent teeth (Figs 1A and B).

Occlusal radiograph indicated multiple radiolucencies around the lateral incisor, canine and first premolar teeth but the cortical bone was intact and no bone expansion was noted (Fig. 2).

Axial CT scans of the mandible (bone-window) revealed well-defined multiple heterogenous hypodense areas with scalloped border that involved incisor, canine and premolar teeth. A slight expansion at the lingual side and eroding of the cortices at buccal side of the tumor were also noted (Figs 3A and B).

The initial diagnosis was a lateral periodontal cyst, keratocystic odontogenic tumor, or small ameloblastoma. Following a tissue biopsy and histopathological examination of the specimen, a diagnosis of ameloblastoma



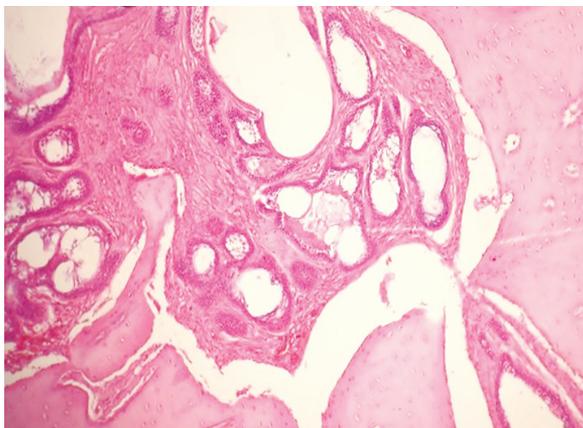
**Figs 3A and B:** (A) Axial computed tomography (CT) scan of mandible (bone-window) shows heterogenous hypodense area approximately 10 mm in diameter and eroding of buccal cortical plate and (B) Axial CT scan (bone window) shows thinning and slight expansion of lingual cortex of the left mandibular corpus and well-defined multilocular hypodense areas surrounding the teeth

was confirmed. The histopathological type of the lesion was follicular type with intraosseous components (Fig. 4).

Surgery consisted of an alveolar segmental resection containing the teeth numbered 31, 32 and 33. For this purpose, a vertical incision was made to the distal papilla of the tooth number 41, and a horizontal incision was extended to the second premolar on the left side. The bone extending from the mesial part of the tooth number 31 to the distal side of the tooth number 33 was demarcated with a saw. To preserve the integrity of the mandibular cortex, the segment was removed as a block, 1 cm apart, from the apical teeth. Histopathological examination of the segment also confirmed the diagnosis of ameloblastoma. However, after a 1-year follow-up, no recurrence was observed.

## DISCUSSION

Our case provides an opportunity to develop a differential diagnosis for radiolucencies associated dental roots, in line with the reports presented by Levin et al<sup>2</sup> and Lagares et al<sup>3</sup> of two cases of small ameloblastomas in the premolar region of mandibles. Moreover, Naitoh et al<sup>6</sup> reported five cases of ameloblastomas detected in the



**Fig. 4:** Epithelial tumor cells, arranged in a palisaded way in the periphery and comprising ameloblast-like cells, are seen. Stellate reticulum-like cells and squamoid cells are observed in the middle (HE40 $\times$ )

interalveolar septum of the teeth located in the incisor-canine mandibular region.

Based on the strength of the histopathological diagnosis, and the clinical and radiographical appearance of the tumor, the case reported by Lagares, et al<sup>3</sup> was classified as the more common solid/multicystic type of ameloblastoma. Although radiography initially suggested a unicystic type of lesion, that diagnosis was rejected because of the lack of cystic findings in the histopathological examination.

Solid/multicystic types of ameloblastoma preferentially affect the mandible. Although unilocular or multilocular images may be seen in both types of ameloblastoma, the former are more common in the unicystic type, while the latter typically indicate the solid/multicystic type.<sup>3</sup>

Initial radiographic examination in the present case revealed a unicystic lesion; however, CT examination showed that the lesion was actually multilocular. This indicated that CT examination, particularly thin cross-sectional examination, is very valuable in the diagnosis of these hard to observe small lesions. The initial (biopsy) and final histopathological examination of the specimen also confirmed the ameloblastoma diagnosis, emphasizing the value of the combination of histopathology and radiology in establishing a diagnosis.

While the molar-ascending ramus is the most prevalent area for the occurrence of multicystic ameloblastomas, a minority of cases are detected in the area between the first premolars, and between the canine and central incisors, as in our case.<sup>1-4</sup> The desmoplastic type of ameloblastoma most commonly occurs in the anterior region and especially in maxilla. The characteristic radiographical feature of this type of tumor is a mixed radiolucent/radiopaque lesion with an ill-defined margin.<sup>7</sup> It is interesting that the lesion in our case had similar clinical

and radiographical features with the desmoplastic type of ameloblastoma, but histopathological examination did not reveal any desmoplastic alterations. The tumor is often asymptomatic. Small lesions can be detected during a routine radiographic examination.<sup>3</sup> In our case, the lesion dimensions were small, and only swelling was evident.

According to our estimates, the lesion was captured at initial stages. Nonetheless, the lesion was rather aggressive, owing to the local aggressive and infiltrative nature of the ameloblastomas.<sup>1,3</sup>

Following a study by Ide, et al<sup>8</sup> which was conducted on 14 cases of unsuspected small ameloblastomas located in the alveolar bone, 60 cases had been reported to date, and only two of these were the periradicular type of lesion seen in our case. Therefore, such lesions appear to be extremely rare. This argument is also supported by the studies of Takeda and Yamamoto,<sup>9</sup> who stated that only four cases of small ameloblastoma located in the alveolar bone had been reported prior to their study. Moreover, Naitoh, et al<sup>6</sup> reported only five cases of ameloblastoma located in the interalveolar septum of the mandibular incisor-canine region.

Ameloblastomas have a common tendency for root resorption and tooth displacement. Some teeth may displace apically.<sup>6</sup> In our case, neither root resorption nor tooth displacement was evident. The relevant teeth also retained their vitality; however, loss of lamina dura and periodontal membrane of the relevant teeth did occur. Some reports have suggested that the desmoplastic type of ameloblastoma develops from the periodontal membrane of the related tooth. In such a case, lamina dura and periodontal ligament of the teeth relevant to the tumor are invisible on radiographs. Recently, Katsura, et al<sup>10</sup> reported a case of desmoplastic ameloblastoma in the canine-premolar region of the right maxilla. The authors speculated that the tumor in their case might have arisen from the periodontal membrane of the relevant teeth, as a radiographical absence of lamina dura and periodontal membrane was noted. Although, the radiographical features of our case were similar, the histopathological examination did not indicate desmoplastic ameloblastoma.

In addition, although the lesion observed in our case was located in the mandible, its border was not easily detectable. This is an interesting point because lesions with ill-defined borders are characteristic of ameloblastomas in the maxilla.<sup>3</sup>

The lesion was initially thought to be a lateral periodontal cyst, a lateral radicular cyst, keratocyst, or ameloblastoma. Lateral periodontal cysts and lateral periapical cysts have a unilocular cystic appearance, similar to that of the present case. They may also reach an appreciable

size, so that swelling may be noted. In a periapical cyst, the related tooth does not respond to electrical pulp testing, whereas in a lateral case, a positive response is registered.<sup>2</sup> As in our case, small odontogenic keratocystic odontogenic tumor are frequently discovered during routine radiographic examinations. However, in our case, unlike in a keratocyst, bone expansion was detected.

The choice of an exact method of ameloblastoma treatment requires assessment of the clinical type (solid/multicystic, unicystic or peripheral), localization, dimension of the tumor, and the age of the patient. For small lesions, simple enucleation may be applied. However, Sampson and Pogrel<sup>5</sup> postulated that curettage of ameloblastoma resulted in unacceptable recurrence rates. The solid/multicystic type is more infiltrative and needs a more aggressive resection, including soft tissues and a 1 to 2 cm wide safety margin.<sup>2-4</sup> Although, the present lesion was a solid/multicystic type, owing to its small dimensions, local excision combined with protection of the inferior cortex was the right therapy choice.

## CONCLUSION

Localization of ameloblastoma in the alveolar bone combined with the small lesion size is an extremely rare condition. In the case reported here, the border and shape of the lesions were indistinguishable from those of a cyst, indicating that tumors of this type might easily be misdiagnosed. Consequently, we confirmed that a combination of CT and histopathological examination is extremely valuable in achieving a correct diagnosis.

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