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Case Report

Acanthomatous ameloblastoma of the mandible: a recurrent odontogenic tumor mimicking multiple cystic lesions: a case report

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ABSTRACT

Ameloblastoma is a benign but locally aggressive odontogenic tumor characterized by a high recurrence rate. Among its histopathological variants, acanthomatous ameloblastoma is uncommon and often poses a diagnostic challenge due to its resemblance to other cystic and neoplastic jaw lesions. We reported a case of recurrent acanthomatous ameloblastoma involving the left posterior mandible in a young adult male with a prior surgical history. The lesion presented as a slowly progressive swelling associated with tooth mobility and pus discharge. Clinical, radiographic, surgical, and histopathological findings were correlated to arrive at a definitive diagnosis. This case highlights the importance of comprehensive evaluation and long-term follow-up in managing recurrent ameloblastomas.

Keywords: Acanthomatous ameloblastoma, Odontogenic tumor, Mandible, Recurrence, Histopathology

INTRODUCTION

Ameloblastoma is a benign odontogenic tumor arising from remnants of the dental lamina, enamel organ, or epithelial lining of odontogenic cysts. Despite its benign nature, ameloblastoma exhibits locally aggressive behavior with a high propensity for recurrence if inadequately treated.[1] The mandible, particularly the posterior region, is the most commonly affected site.[2]

The acanthomatous variant is a rare histopathological subtype characterized by extensive squamous metaplasia and keratin formation within the stellate reticulum-like cells.[3] Clinically and radiographically, this variant may mimic other odontogenic cysts and tumors, making diagnosis challenging. This report presents a recurrent case of acanthomatous ameloblastoma of the mandible with emphasis on clinicopathological correlation and differential diagnosis.

CASE REPORT

Chief complaint

A 30-year-old male patient reported with a complaint of swelling in the lower left back tooth region for the past one year.

History of present illness

The patient was apparently asymptomatic one year prior when he first noticed a small swelling associated with mobility of teeth in the lower left posterior mandibular region (Figure 1).

The swelling gradually increased in size and was associated with intermittent pus discharge. The patient had undergone surgical intervention for a similar lesion two years earlier, following which he was symptom-free for a period. Recurrence of swelling (Figure 2) at the same site

prompted him to report to the Department of Oral Medicine and Radiology, Subharti Dental College and Hospital, Meerut.

Past medical and dental history

The patient's medical history was unremarkable, with no reported systemic illnesses. There was no history of previous dental treatment or any significant dental pathology.

Personal history

The patient demonstrated inadequate oral hygiene practices, brushing only once daily using a horizontal technique with a soft-bristled toothbrush and herbal toothpaste. Habit history revealed long-standing use of smokeless tobacco (Nevla), approximately one packet every two to three days for the past five years, along with frequent alcohol consumption.

Clinical examination

Extraoral examination

A diffuse swelling was observed on the left side of the mandible, extending posteriorly toward the angle of the mandible. The overlying skin appeared normal with no visible discoloration or ulceration. On palpation, the swelling was hard, tender, non-mobile, and not fixed to the overlying skin, involving the inferior border of the mandible.

Intraoral examination

Soft tissue findings

A large, diffuse expansile swelling was noted in the left buccal vestibule, extending from the region of the mandibular left canine to the second molar, with evident buccal cortical plate expansion. The overlying mucosa appeared pink and intact. The gingiva, palate, floor of the mouth, and tongue showed no abnormalities.

Hard tissue findings

Teeth 35 and 36 were missing. The patient exhibited a bilateral Class I canine relationship. Periodontal examination revealed moderate calculus deposits with no bleeding on probing and no mobility of the remaining teeth.

The swelling measured approximately 3-4 cm in diameter and was firm and tender on palpation.

Provisional diagnosis

Based on the clinical findings, a provisional diagnosis of an oontogenic tumor was made.

Differential diagnosis

Considering the size, location, and clinical behavior of the lesion, the differential diagnosis included odontogenic keratocyst, central giant cell granuloma, dentigerous cyst, odontogenic myxoma, calcifying odontogenic cyst, calcifying epithelial odontogenic tumor, aneurysmal bone cyst, and fibrous dysplasia.

Radiographic findings

Panoramic radiograph (Figure 3) revealed a multilocular radiolucent lesion involving the left posterior mandible extending from the premolar to molar region, with evidence of cortical expansion and displacement of adjacent structures, suggestive of an aggressive odontogenic lesion.

Surgical management

The lesion was surgically enucleated (Figure 4) and curetted under aseptic conditions. Multiple soft and hard tissue fragments were obtained and submitted for histopathological examination.

Histopathological examination

Hematoxylin and eosin- stained sections demonstrated multiple odontogenic epithelial follicles (Figure 5). These follicles were lined peripherally by tall columnar ameloblast-like cells arranged in a palisaded pattern, with nuclei polarized away from the basement membrane. The central portion of the follicles consisted of loosely arranged stellate reticulum–like cells exhibiting areas of squamous metaplasia. Foci of individual cell keratinization and keratin pearl formation were evident within the epithelial islands. The surrounding stroma showed marked vascularity with areas of hemorrhage. These microscopic features were consistent with a diagnosis of acanthomatous ameloblastoma.

Final diagnosis

Acanthomatous ameloblastoma of the mandible.

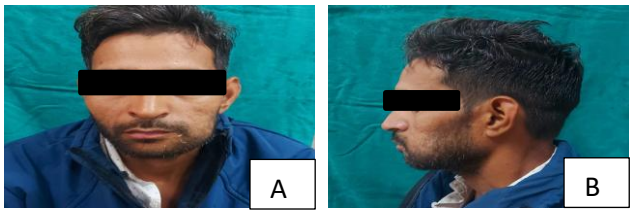

Figure 1: Pre-operative extraoral clinical photographs showing facial asymmetry due to swelling involving the left posterior mandible (A) frontal view demonstrating mild facial fullness on the left side; and (B) lateral profile view showing anteroposterior extension of the swelling up to the angle of the mandible with normal overlying skin.

Figure 2: Pre-operative intraoral clinical photographs (A) intra-oral view using cheek retractors revealing a diffuse expansile swelling involving the left mandibular posterior region (33-38) with intact overlying mucosa; and (B) closer intraoral view demonstrating buccal cortical expansion with erythematous areas suggestive of underlying pathology.

Figure 3: Panoramic radiograph (orthopantomogram) showing a multilocular radiolucent lesion involving the left posterior mandible extending from the premolar to molar region, associated with cortical expansion and missing teeth (35 and 36), suggestive of an aggressive odontogenic lesion.

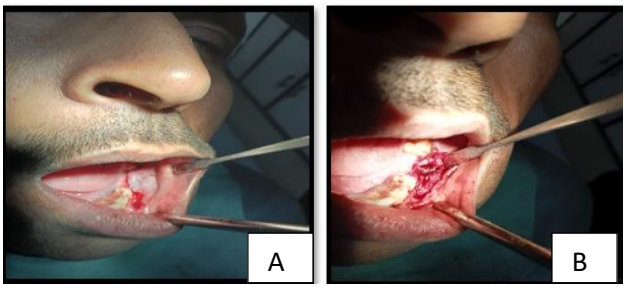

Figure 4: Intra-operative photographs (A) surgical exposure of the lesion following mucoperiosteal flap reflection, revealing extensive bone involvement; and (B) intra-operative view showing curettage and removal of pathological tissue from the left posterior mandible.

DISCUSSION

Ameloblastoma is a benign but locally aggressive odontogenic neoplasm that accounts for approximately 1% of all oral tumors and about 10–15% of odontogenic tumors reported in the literature.[1] Among its histopathological variants, the acanthomatous type is characterized by prominent squamous metaplasia within the stellate reticulum, often accompanied by keratin pearl formation, as observed in the present case. These histologic features are in agreement with the descriptions provided by Reichart et al. and Neville et al., who emphasized that squamous transformation is a defining feature of the acanthomatous variant and can occasionally mimic well-differentiated squamous cell carcinoma if epithelial atypia is not carefully evaluated.[2]

The age of the patient in the present case (30 years) falls within the commonly reported age range for ameloblastoma, which predominantly affects individuals between the third and fifth decades of life. Several large retrospective studies have demonstrated a peak incidence in the third decade, supporting the demographic profile of the current case. A slight male predilection has been reported in some populations, which further correlates with the present finding of a male patient. Anatomically, the posterior mandible is the most frequent site of involvement for ameloblastoma, particularly in the molar-ramus region. This predilection has been attributed to the persistence of odontogenic epithelial remnants, especially the dental lamina and enamel organ, in this region.[4] The lesion in the present case involved the left posterior mandible extending from the premolar to molar region, consistent with the location reported in the majority of published series. Clinically, ameloblastomas typically present as slow-growing, painless swellings that may remain asymptomatic for long periods before becoming noticeable due to facial asymmetry or tooth displacement. However, secondary infection may result in pain and pus discharge, as seen in the present patient. Similar findings have been reported by Small and Waldron, who noted that although ameloblastomas are generally painless, superimposed infection can produce tenderness and suppuration. The presence of tooth mobility in this case is also in accordance with previous studies describing alveolar bone destruction and periodontal ligament involvement.

Radiographically, ameloblastomas most commonly present as multilocular radiolucencies with a ‘soap bubble’ or ‘honeycomb’ appearance.[5] The panoramic radiograph in the present case revealed a multilocular radiolucent lesion with cortical expansion and tooth loss, which closely matches the radiographic patterns described by Kahn and by Hong et al.[6] These features reflect the infiltrative and expansile nature of the tumor. Histopathologically, the present lesion demonstrated odontogenic epithelial follicles with peripheral palisading of tall columnar ameloblast-like cells and central stellate reticulum-like cells showing extensive squamous metaplasia and keratin

pearl formation.[7] These findings are diagnostic of acanthomatous ameloblastoma and are consistent with those reported by Philipsen and Reichart, who highlighted that this variant shows prominent keratinization within epithelial islands.

The recurrence observed in this patient is an important clinical feature. Recurrence is a well-documented characteristic of ameloblastoma, particularly when treated conservatively by enucleation or curettage. Studies by Nakamura et al. and Lau and Samman have demonstrated significantly higher recurrence rates following conservative surgical approaches compared to marginal or segmental resection.[8] The recurrence in the present case is likely attributable to residual microscopic tumor islands left behind during the previous surgical procedure, which is a known limitation of conservative management. From a prognostic standpoint, acanthomatous ameloblastoma does not differ significantly from other solid multicystic variants in terms of biological behavior; however, its infiltrative growth pattern necessitates careful surgical planning and long-term surveillance. Long-term follow-up is strongly recommended, as recurrences have been reported even 10-15 years after initial treatment. In summary, the clinical, radiographic, and histopathological features of the present case closely parallel those described in earlier studies on acanthomatous ameloblastoma. The recurrence underscores the importance of adequate surgical margins and prolonged follow-up in managing this aggressive odontogenic tumor.

CONCLUSION

Acanthomatous ameloblastoma, though benign, demonstrates aggressive local behavior and a high tendency for recurrence. Accurate diagnosis requires careful clinicoradiographic correlation supported by histopathology. Adequate surgical management and prolonged follow-up are essential to prevent recurrence and associated morbidity.

Clinical significance

Early diagnosis and definitive treatment planning are crucial in recurrent ameloblastomas to reduce patient morbidity and improve prognosis.

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