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A case report

Slow Growth Mixed-Type Ameloblastoma of Anterior Mandible in a Middle-Age Patient

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Abstract:

Introduction: Ameloblastoma is the most common odontogenic tumor, with an annual global incidence of 0,5 cases per 1.000.000 people. Histologically it can be follicular, plexiform, desmoplastic, and acanthomatous. Mixed-type ameloblatomas show a combination of these subtypes.

Case report: This report presents the case of a slow-growing mixed-type ameloblastoma in the left mandible of a 51-year-old male patient. Diagnosis was established based on clinical, radiological, and histopathological examinations. Segmental resection with adequate margins and AO plate reconstruction was performed. Postoperative histopathological examination confirmed mixed-type ameloblastoma with clear resection margins.

Discussion: In this case, the patient had a slow-growing mixed-type ameloblastoma of the mandible. From the histopathological examination shows a tumor mass consisting of hyperplastic odontogenic epithelial cells, grouped together to form a partially plexiform and partially acanthomatous structure which is particularly a rare characteristic of mixed-type ameloblastoma. In addition to the plexiform and acanthomatous structures, some of the cells were squamous differentiated with palisading nuclei at the edges, characterizing acanthomatous subtype of ameloblastoma.

Conclusion: There are only a few reports presenting ameloblastomas mixed-type especially involving the acanthomatous subtype, further research is needed on the factors that influence the occurrence of mixed-type ameloblastoma.

Keywords: Ameloblastoma, Mandible, Mixed-type ameloblastoma, Acanthomatous, Plexiform

Introduction

Ameloblastoma is one of the most benign tumor originating from tooth-forming tissues, occurs at a global rate of about 0.5 cases per million people annually, with significant geographic variation. [1,2] This uncommon growth primarily develops from undifferentiated enamel tissue.[1,3] While benign, ameloblastoma exhibits locally aggressive behavior, growing slowly and often leading to delayed diagnosis.[4]

Initially, the lesion is typically asymptomatic, but as it progresses, individuals may experience pain, loose teeth, facial asymmetry, and difficulty speaking.[1] Ameloblastoma most frequently develops in the mandible (70%-80%), mainly in the molar region and ascending ramus (70%), followed by the premolar (20%), and anterior region (10%). Tumors may be arisen in the maxilla (20%), predominantly in the canine and molar region. Sometimes, an unerupted third molar can be associated the development of ameloblastoma.[3]

Ameloblastoma typically appear as radiolucent areas on radiographs, presenting either as unilocular or muoltilocular lesions (soap bubble). Although root resorption is uncommon, it can occur, particularly in fast-growing tumors. [5] The current standard of care involves radical surgical resection with a minimum safety margin of 1 cm. Recurrence rates following such radical treatment range from 0% to 15%.1 In contemporary practice, wide resection followed by immediate reconstruction is considered the treatment of choice for most cases of mandibular ameloblastoma. However, recurrence can still occur, with rates reported between 15-25% even after radical treatment and 75-90% after conservative treatment. Delayed diagnosis and management can significantly contribute to increased morbidity, particularly in the form of severe facial disfigurement and compromised oral function.[4]

Ameloblastoma is classified into four main types: conventional ameloblastoma, unicystic ameloblastoma, extraosseous or peripheral ameloblastoma, and metastasizing (malignant) ameloblastoma.[3] Several histopathological subtypes have been identified, with the follicular variant being the most common, accounting for approximately 64.9% of cases. This is followed by the plexiform subtype (13%), desmoplastic subtype (5.2%), and the acanthomatous variant (3.9%). In addition, mixed-type ameloblastomas exhibit a combination of these histological patterns within a single lesion.[2]

The objective of this article is to report our experience with treating an interesting case of mixed-pattern ameloblastoma, including components of both the plexiform and acanthomatous subtype, located at the anterior mandible.

Case Presentation

A 52 years old male presented with a chief complaint of a large, painless mass in the anterior mandible, progressively enlarging over 16 years. The lesion began as a small, soft swelling two years after a molar extraction. Temporary regression was noted following empirical medications, but the mass gradually expanded to a softball-sized lesion, causing significant facial asymmetry.

The patient denied pain, bleeding, or systemic illness, but reported unintentional weight loss of 30 kg in the past year. Extraoral examination (Figure 1,2) revealed a dome-shaped mass spanning from the left to right parasymphysis, with focal ulceration and crusting. Imaging (Figure 3,4,5) showed a large, multilocular, soap bubble-like radiolucency involving the left to anterior mandible, with cortical expansion and soft tissue involvement. An incisional biopsy confirmed a diagnosis of follicular ameloblastoma. Panoramic radiograph (Figure 4) was performed and it showed a defect along the left mandibular, a soap bubble with mixed radiolucent and radio-opaque appearance extending from 37 to 45 region to the inferior border of the mandible. A CT scan examination (Figure 5) was performed and revealed a multicystic expansile mass exhibiting a characteristic “soap bubble appearance,” seemingly originating from the left mandibular ramus. The lesion was seen obliterating the buccinator muscle, mentalis muscle, depressor labii inferior muscle, and

depressor anguli oris muscle. The lesion also causing destruction of the mandibular body.

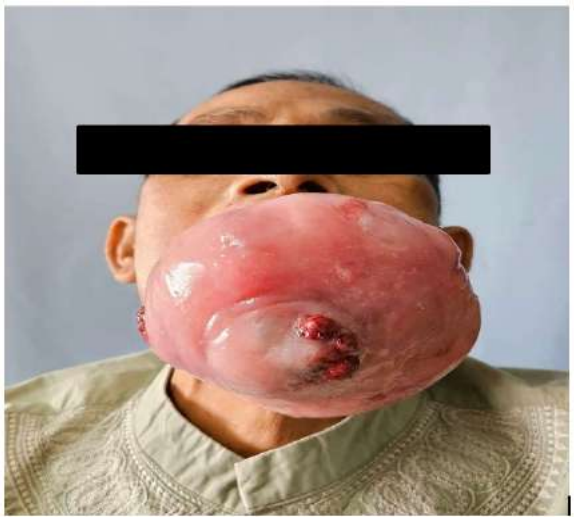

Figure 1: Patient profile condition

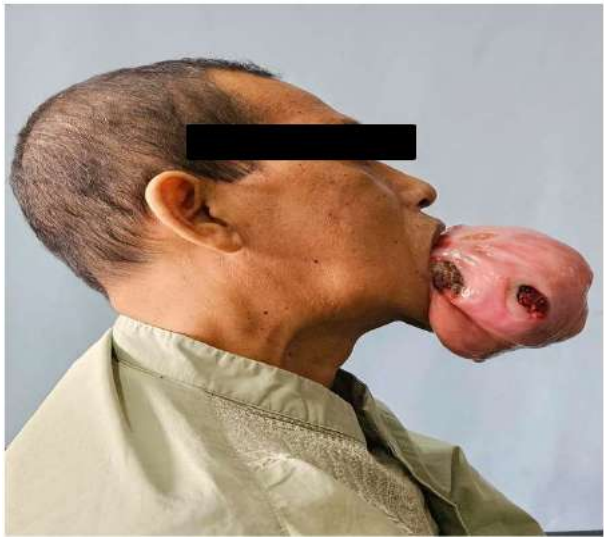

Figure 2: Patient left lateral profile

Figure 3: Intraoral examination.

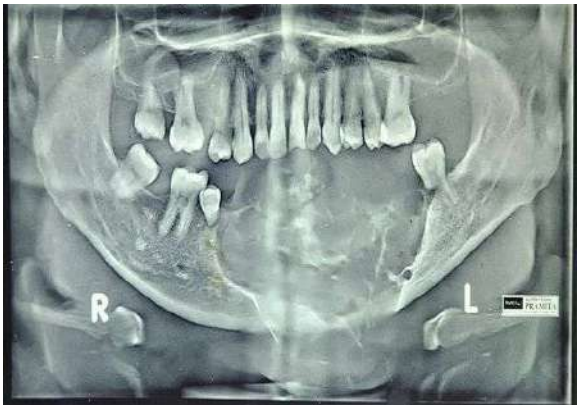

Figure 4: Panoramic radiograph

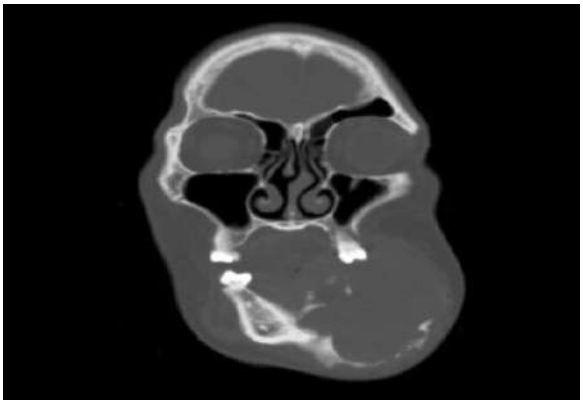

Figure 5: Coronal CT Scan examination

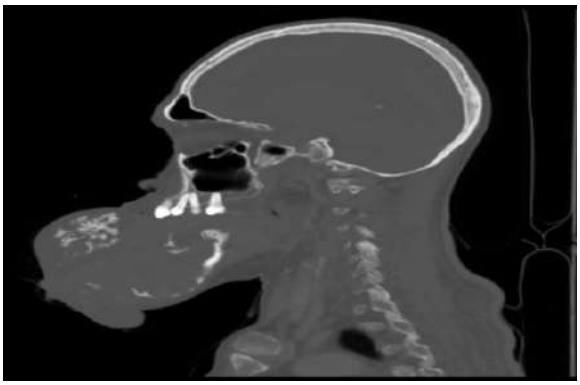

Figure 6: Lateral CT Scan examination

The patient subsequently underwent segmental resection (Figure 6) under general anesthesia with safe margins. The specimen (Figure 7) measured 15 × 12 × 7 cm and was histopathologically confirmed as a mixed-pattern ameloblastoma (plexiform and acanthomatous types). An AO reconstruction plate (Figure 8) was placed, and the wound was closed in layers (Figure 9). Histopathological examination of the resected specimen revealed that the definitive diagnosis was mixed-pattern ameloblastoma, characterized by the presence of both plexiform and acanthomatous components. A follow-up and suture removal was performed several weeks after surgery, and the patient reported no complaints

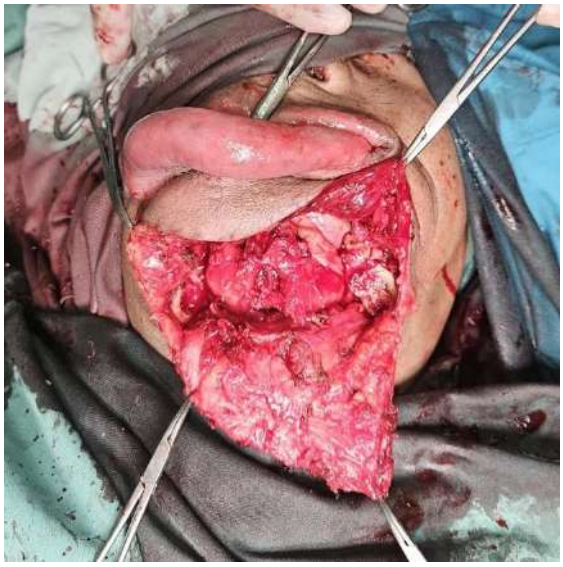

Figure 7: A segmental resection was perfomed under general anesthesia

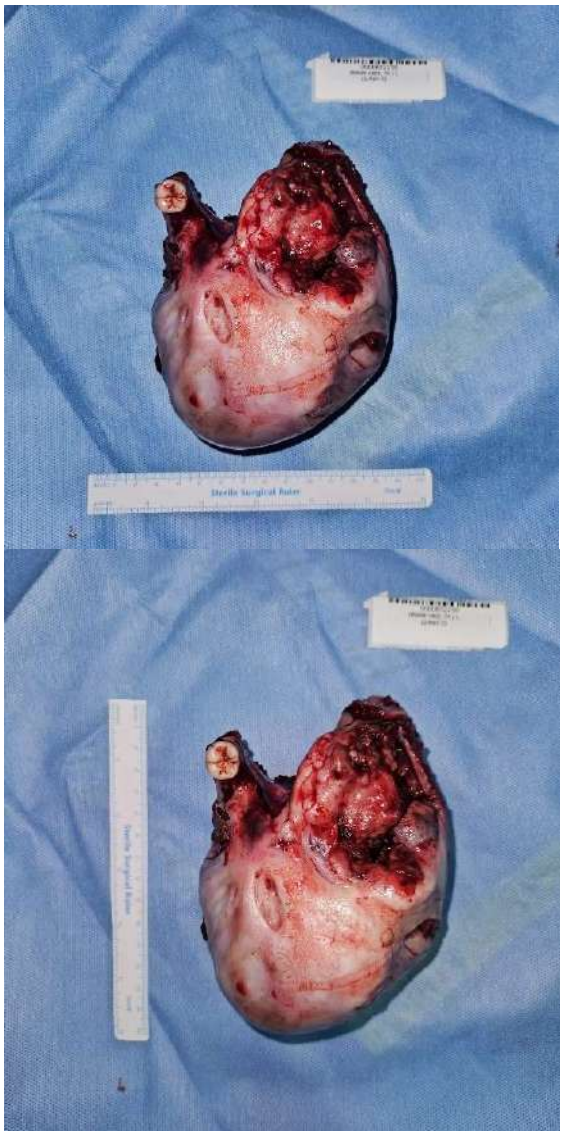

Figure 8: A resected specimen

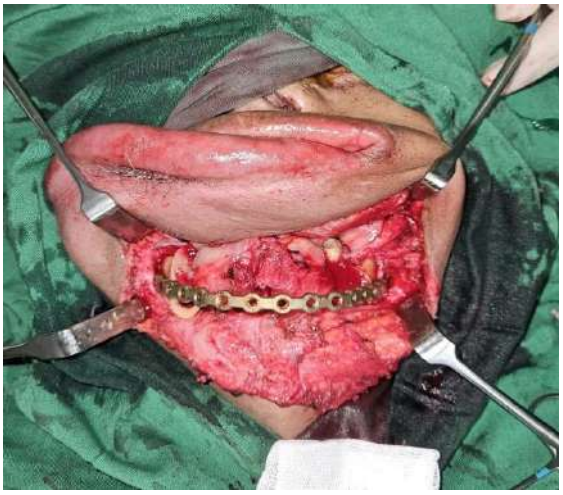

Figure 9: An AO plate placed on the left mandible

Figure 10: Closed surgical wound

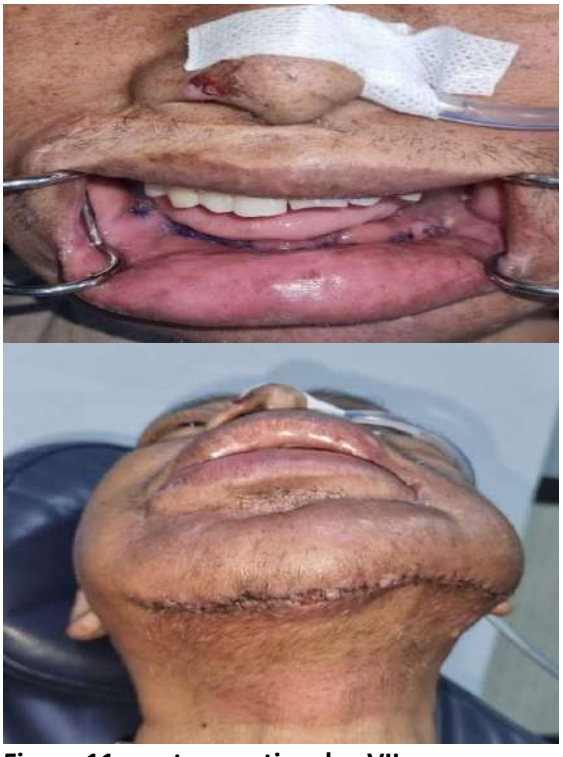

Figure 11: post-operative day VII

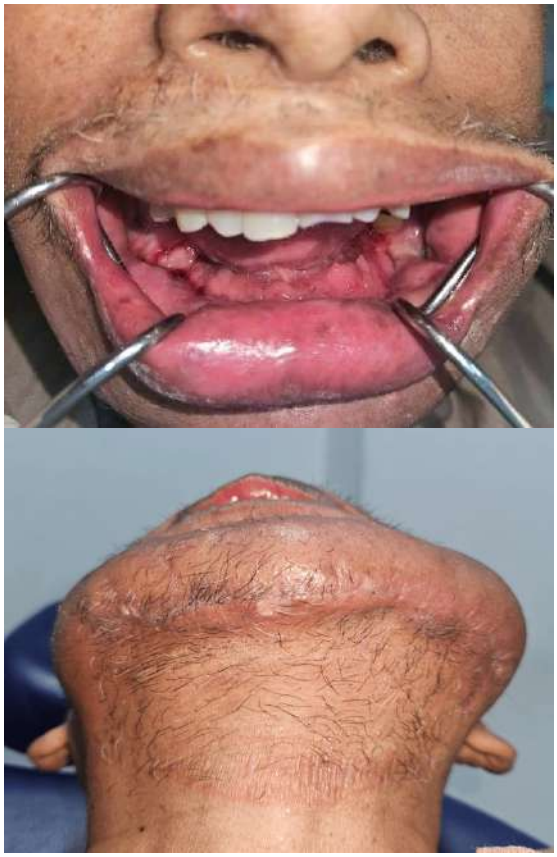

Figure 12: Post-operative day XIV

Discussion

Ameloblastoma is the most common odontogenic tumor affecting the jaws, accounting for approximately 48.9% of all odontogenic neoplasms and 9% of all epithelial odontogenic lesions. It is considered a true neoplasm of the enamel organ, characterized by persistent local growth, a high tendency for locoregional invasion, and a non-encapsulated nature. Although benign, it rarely metastasizes and does not undergo differentiation till the point it is capable of forming hard tissue (enamel). [6]

According to the World Health Organization (WHO), ameloblastomas can be classified as solid/multicystic, unicystic, extraosseous/peripheral, and desmoplastic. [7,8] Histologically, they are further categorized into six subtypes: follicular, plexiform, desmoplastic, acanthomatous, basal cell, and granular cell types. Among these, the follicular subtype is the most prevalent (64.9%), followed by plexiform (13%), desmoplastic (5.2%), and acanthomatous (3.9%) variants. [9] Microscopically, ameloblastomas have a fibrous stroma with islands or masses of proliferating epithelium resembling odontogenic epithelium of the enamel organ and palisading of cells around proliferating epithelium in a pattern similar to ameloblasts. [6–8]

Mixed-type ameloblastomas, which demonstrate more than one histological subtype within a single lesion, are relatively uncommon. Although ameloblastoma with more than one epithelial histo-type are not infrequent, but mixed cases with acanthomatous hysto-type are rarely reported in the literature. [9] The present case exhibited histopathological features of both acanthomatous and plexiform types, which is particularly rare and aligns with few reported cases by Hertog et al., Jain et al., and Figueiredo et al., where mixed histological patterns were identified in single tumors. [7,8]

The acanthomatous variant is characterized microscopically by central squamous cell differential with keratin formation. [6–8] It most commonly occurs in the posterior mandible of older adults, with a peak incidence around the seventh decade of life (mean age: 61.3 ± 1.2 years). [2,6,8] Acanthomatous metaplasia has been associated with chronic irritation, often resulting from dental calculus and oral sepsis. [2,8] Similarly, in this case, the patient experienced chronic irritation at the site of a previous dental extraction that remained untreated for over a decade, which may have contributed to the acanthomatous transformation.

Plexiform ameloblastoma, typically seen in the fifth to sixth decades of life, is most commonly found in the posterior mandible. Histologically, it is characterized by cells arranged in interconnecting strands and cords with cuboidal or columnar basal cells exhibiting hyperchromatic nuclei, nuclear palisading with polarization and cental stellate reticulum like-cells. [6,8] In the

current case, such plexiform patterns were clearly visible alongside acanthomatous features on histological examination.

Radiographically, this case initially resembled a unicystic ameloblastoma, a form often seen as a unilocular radiolucency. However, the CT scan revealed a multicystic expansile lesion with a classic "soap bubble" appearance, causing destruction of the mandibular body and infiltration into adjacent soft tissues, including the buccinator, mentalis, depressor labii inferior, and depressor anguli oris muscles. This radiographic presentation is consistent with previous studies showing cortical and soft-tissue destruction due to infiltration of tumor cells, especially into the cancellous portion of the cortical bone.[8]

The management of ameloblastomas requires careful consideration of multiple factors, including the clinico-radiologic subtype, anatomical location, tumor size, biological behavior, and the patient's overall condition. Conventional ameloblastomas are typically locally invasive and exhibit a high recurrence rate if not excised adequately. The choice between conservative and radical surgical approaches must be tailored to the tumor's macroscopic and histological characteristics. Conservative treatments—such as enucleation, curettage, or limited surgical excision—are often reserved for smaller, unicystic or unilocular lesions but are associated with significantly higher recurrence rates, ranging from 60% to 90%. In contrast, radical approaches, including marginal or segmental resection with 1–2 cm of uninvolved bone, have demonstrated lower recurrence rates, albeit at the expense of increased morbidity, as well as potential functional and cosmetic deficits. For extensive lesions, especially those compromising a significant portion of cortical bone or adjacent anatomical structures, more aggressive management is necessary, including supra-periosteal resection and soft tissue removal. In the present case, the lesion's size and involvement of the mandibular body necessitated a segmental resection with AO plate reconstruction. This decision was made following thorough clinical evaluation, as tumor growth had stabilized and surgical morbidity was deemed acceptable. In contrast, conservative management is generally favored in younger patients to minimize developmental and functional impairment. [1,7]

Conclusion

This case highlights a rare presentation of mixed-type ameloblastoma involving both acanthomatous and plexiform subtypes, located in the anterior mandible. The patient's prolonged clinical course and the tumor's slow progression underscore the locally aggressive nature of ameloblastomas despite their benign classification. Histopathological confirmation of multiple subtypes within a single lesion emphasizes the importance of thorough diagnostic evaluation in guiding appropriate management. Segmental resection with adequate margins remains the definitive treatment for extensive lesions, offering favorable outcomes and minimizing recurrence. Given the rarity of mixed-type variants, particularly those with acanthomatous features, further studies are warranted to better understand their pathogenesis, clinical behavior, and optimal management strategies.

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