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Successful surgical management of acanthomatous ameloblastoma with extensive mandibular involvement; a rare case report

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Abstract

Ameloblastoma is the most common benign odontogenic epithelial tumor, accounting for approximately 11–18% of all odontogenic neoplasms and 1-3% of all oral cysts and tumors. It primarily affects the posterior mandible and usually arises in the third to fifth decades of life. Despite its slow growth, this tumor is locally aggressive and may metastasize or undergo malignant transformation. Histologically, ameloblastoma comprises several variants. The acanthomatous variant, although relatively rare, is characterized by squamous metaplasia and keratin formation within the central stellate reticulum-like cells of the tumor islands. The clinical and radiographic characteristics of ameloblastoma are often nonspecific and may resemble other multilocular radiolucent lesions. Accurate diagnosis necessitates the integration of advanced radiographic imaging, particularly cone-beam computed tomography, along with definitive histopathologic evaluation. We presented a rare case of acanthomatous ameloblastoma affecting both the anterior and posterior regions of the left mandible in a 33-year-old male with a five-year history of jaw expansion and progressive tooth mobility, along with its surgical management. The lesion showed extensive anterior extension, crossing the midline. This case underscores the significance of comprehensive differential diagnosis and histopathological confirmation in managing unusual presentations of ameloblastoma.

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Introduction

Ameloblastoma is recognized as the most common benign odontogenic tumor (1). Clinically, it has potentially aggressive, painless, and slow-growing characteristics and typically remains asymptomatic (2-4). There is a high potential for malignant transformation as well as metastasis in ameloblastoma lesions (5). However, ameloblastoma more commonly presents with a high recurrence rate rather than metastasis (2). Ameloblastoma represents 11-18% of odontogenic tumors and 1-3 % of all oral tumors and cysts, with no gender predilection (5-8). Solid/ multicystic, unicystic, desmoplastic, and extraosseous/ peripheral are the four major histopathological types of ameloblastoma introduced by WHO in 2005 (9). Follicular, plexiform, granular, basal, desmoplastic, and acanthomatous are among the histological subtypes described for ameloblastoma (2-4,7).

The term acanthomatous denotes the presence of extensive squamous metaplasia and variable keratinization within stellate reticulum-like cells. In addition, the formation of squamous islands within the central regions of the neoplastic nests, as well as areas of calcification, may be observed. Acanthomatous ameloblastoma exhibits histopathological features closely resembling those of the squamous odontogenic tumor, from which it is distinguished by the columnar morphology of the peripheral cells rather than a flattened configuration (9). This report presents a rare case of acanthomatous ameloblastoma affecting both anterior and posterior regions of the mandible in a 33-year-old male, with a five-year history of progressive jaw swelling and tooth mobility.

Case Presentation

A 33-year-old male was referred to the oral and maxillofacial surgery department with complaints of unilateral facial swelling and

Key point

Ameloblastoma is a locally aggressive benign odontogenic tumor, with the rare acanthomatous variant showing squamous metaplasia and keratin formation. We report a rare case in a 33-year-old male with extensive mandibular involvement crossing the midline. This case emphasizes the need for advanced imaging and histopathological confirmation to ensure accurate diagnosis and effective management.

increased mobility of the anterior mandibular teeth. The patient reported that the lesion and associated swelling had been present for approximately five years, during which he had declined treatment. His medical history was unremarkable, and he did not report any systemic conditions. Although the patient denied pain, he occasionally experienced paresthesia in the affected region.

Extraoral examination revealed a prominent unilateral facial swelling localized to the left side of the mandible, extending anteriorly (Figure 1).

Intraoral evaluation demonstrated diffuse swelling in the anterior mandibular region, with significant buccal and lingual expansion of the left posterior mandible (Figure 2). Displacement of multiple teeth, particularly the left lateral incisor, canine, and first premolar, was observed.

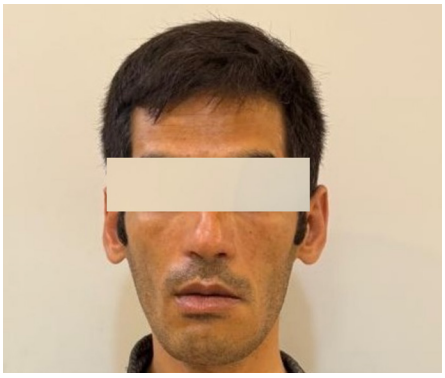

Figure 1. Extraoral photograph showing facial swelling localized to the left side of the mandible.

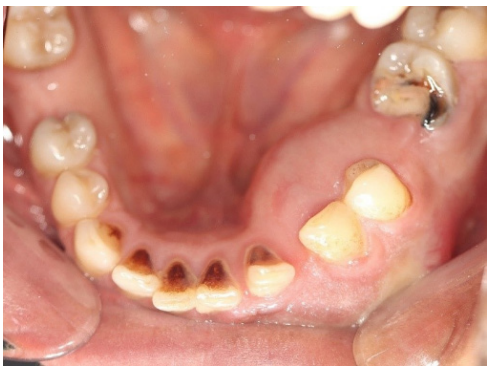

Figure 2. Intraoral photograph demonstrating buccal and lingual expansion of the left posterior mandible.

The overlying mucosa appeared pink and intact, without evidence of ulceration, fistula, or bleeding.

Radiographic assessment

An initial panoramic radiograph was obtained using a digital imaging device (PaX-i Plus; Vatech, South Korea), which revealed a well-defined, multilocular, radiolucent lesion involving the left mandibular body and extending toward the anterior region (Figure 3).

To comprehensively assess the lesion's size, extent, and relationship to adjacent structures, cone-beam computed tomography (CBCT) was performed using a CBCT imaging unit (Giano HR; NewTom, Italy) with a mandibular field of view. Multiplanar reconstructions (axial, coronal, sagittal, and cross-sectional views) were evaluated using NNT Viewer software. Meanwhile, CBCT imaging demonstrated a large, expansile, multilocular radiolucent lesion occupying the left mandibular body and extending across the midline to the region of the right canine (Figure 4). The lesion spanned horizontally from the periapical area of the left first molar to the right mandibular canine and vertically from the alveolar crest to the inferior mandibular border. The borders were well-defined and corticated, with areas exhibiting scalloping. The internal structure displayed coarse, thick, and

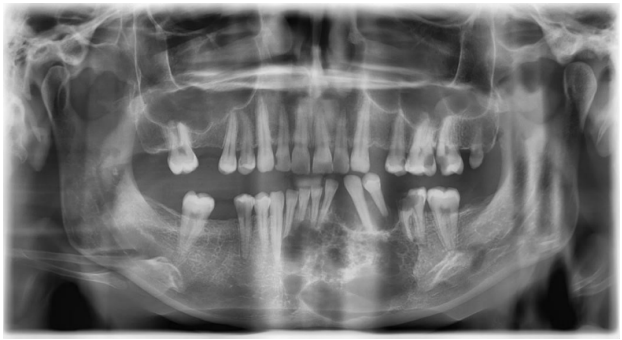

Figure 3. Panoramic radiograph revealing an expansile multilocular radiolucency involving the anterior and left mandibular regions.

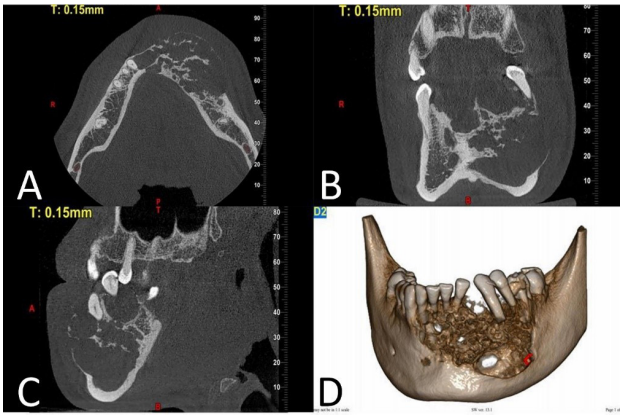

Figure 4. CBCT images: A) axial view; B) coronal view; C) sagittal view; D) three-dimensional reconstruction. Note the bony perforation, honeycomb appearance, tooth displacement, and cortical expansion.

curved septa, producing a characteristic "soap-bubble" and "honeycomb" radiographic appearance. Significant thinning, outward expansion, and focal perforation of both buccal and lingual cortical plates were observed. The lesion caused root resorption of several involved teeth and displacement of adjacent dentition. Destruction of the lamina dura and widening of the periodontal ligament space were also evident.

Based on the clinical and radiographic findings—including extensive jaw expansion, root resorption, cortical destruction, and a multilocular internal pattern—the primary radiographic differential diagnoses included ameloblastoma, central giant cell granuloma, and odontogenic keratocyst (OKC). The patient was subsequently referred for surgical excision and incisional biopsy.

Histopathological evaluation

A biopsy was conducted several weeks prior to definitive surgical treatment. The histopathology specimen, received in formalin, consisted of multiple soft, creamy-tan fragments, collectively measuring 2×1.5×0.6 cm.

Microscopic examination of hematoxylin and eosin (H&E)-stained sections revealed odontogenic epithelial islands composed of columnar cells with hyperchromatic nuclei at the basal layer, arranged in a palisading pattern. These cells exhibited reverse nuclear polarization away from the basement membrane (Vickers-Gorlin change) and sub-nuclear vacuolization. The supra-basal cells displayed a loose, network-like configuration, recapitulating stellate reticulum. Extensive squamous metaplasia with keratin formation within the central stellate reticulum-like cells of the epithelial islands was observed. No dentin or enamel formation was identified. The odontogenic epithelial islands and nests were separated by mature fibrous stroma (Figure 5). Histopathological evaluation was consistent with the acanthomatous variant of ameloblastoma.

To further characterize the tumor, immunohistochemical (IHC) staining was performed using a panel of markers. The neoplastic epithelial cells showed positive immunoreactivity for CK19 and CK14, confirming odontogenic epithelial origin. β-catenin showed strong membranous and focal cytoplasmic positivity, indicating active epithelial proliferation. In addition, BRAF V600E mutation testing was positive, supporting its role in the pathogenesis of ameloblastoma and suggesting potential implications for targeted therapy (Figure 6).

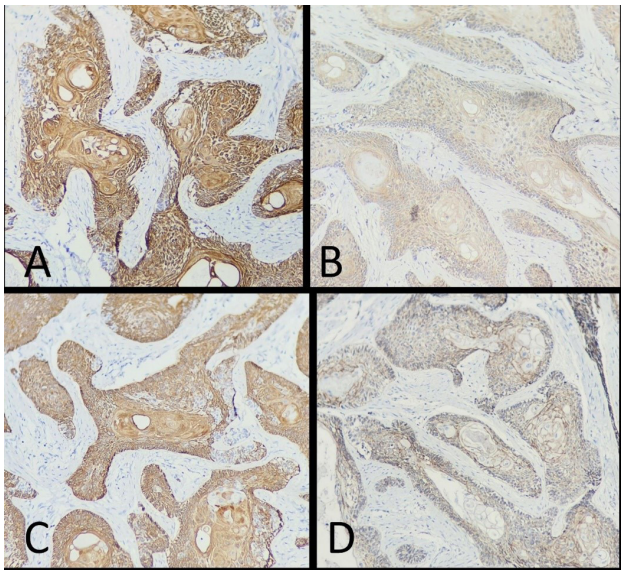

Figure 6. Immunohistochemical staining: A) CK19 highlights tumor cells in both peripheral and stellate reticulum-like areas; B) BRAF V600E shows positivity in tumor cells within the peripheral and stellate reticulum-like areas; C) CK14 labels tumor cells in the peripheral and stellate reticulum-like areas; D)β-catenin expression is increased with membranous localization (×200).

Surgical treatment

Following histopathological evaluation and confirmation of the diagnosis, the patient was admitted for surgical management. Informed consent was obtained from the patient. A segmental mandibular resection was performed via an extraoral approach under general anesthesia, maintaining a 1cm oncologic safety margin. The resulting discontinuity defect was reconstructed using a patient-specific custom reconstruction plate with a meshed crib design (Figure 7).

Figure 8 demonstrates the surgical process. The plate was covered with muscular flaps derived from the tongue, floor of the mouth, and platysma to reduce the risk of

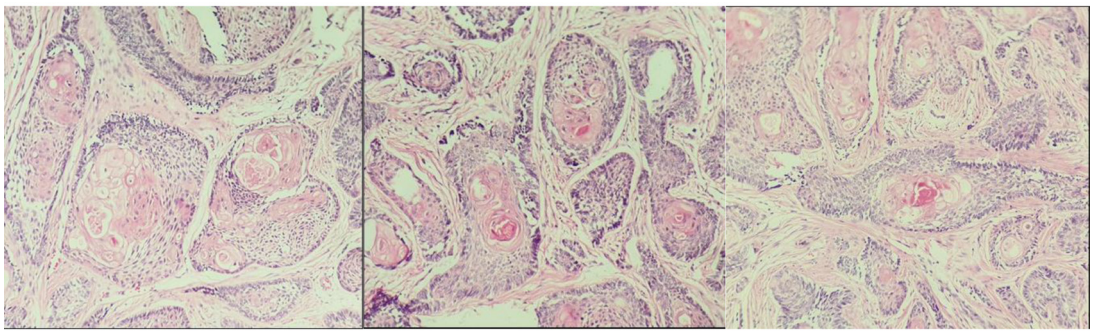

Figure 5. Histopathological section showing odontogenic epithelial islands with peripheral palisading columnar basal cells and central stellate reticulum. Peripheral basal cells exhibit reverse nuclear polarity. Extensive squamous metaplasia with keratin formation is evident within the stellate reticulum-like cells in the central portion of the epithelial islands (×200).

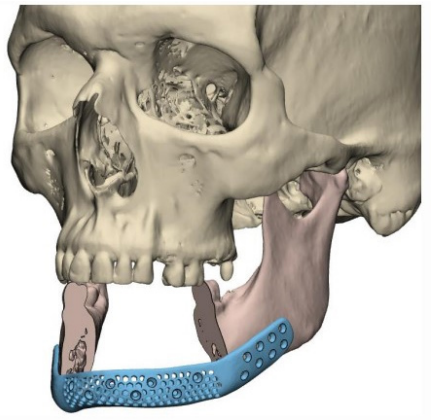

Figure 7. Patient-specific custom reconstruction plate with a meshed crib design.

wound dehiscence.

The patient was scheduled for postoperative follow-up at 1 week, 1 month, and 3 months. Healing progressed uneventfully, with no clinical evidence of infection or wound-related complications (Figure 9).

Once recurrence of the primary tumor has been definitively excluded, the patient will undergo secondary reconstruction using a particulate autogenous bone graft placed within the existing custom-designed reconstruction plate.

Discussion

Ameloblastoma, a relatively rare odontogenic neoplasm, has been described as the most common odontogenic tumor and is one of the most controversial tumors of the facial skeleton due to the presence of various classifications and the lack of standardization among published studies (1,10). An extensive review article comprising 3,677 cases reported the mean age at initial diagnosis of ameloblastoma as 35.9 years, with older ages noted for the acanthomatous variant (10). In general, the peak incidence of ameloblastoma occurs between the third and fifth decades of life, which is consistent with the case presented in this report (2). However, Adebiyi et al (11) reported the seventh decade of life as the period of highest prevalence for acanthomatous ameloblastoma. This discrepancy may be attributed to the small number of acanthomatous ameloblastoma cases included in their study, as the diagnosis was made in only three patients. Furthermore, their study was conducted exclusively in Nigerian patients, and differences among population groups may have influenced their findings.

Previous literature has reported that clinical symptoms of ameloblastoma may precede the initial diagnosis by 2 to 6 years (10). Similarly, our patient reported a history of swelling for approximately five years. The patient recalled attending a dental appointment several years earlier for evaluation of the swelling, during which a fine-needle aspiration biopsy was performed. At that time, a diagnosis of OKC was rendered. Despite the diagnosis, the patient declined further treatment, and the lesion remained untreated. The likely cause of this misdiagnosis was the abundant keratin content within the ameloblastoma and the reliance on a fine-needle aspiration specimen rather than a representative tissue biopsy. However, in their literature review, Kumar et al (12) reported nine cases of ameloblastoma arising from the epithelial lining of OKC.

Although ameloblastoma is frequently asymptomatic, patients may present with jaw expansion, tooth displacement, malocclusion, tooth mobility, facial swelling, paresthesia, anesthesia, and occasionally pain (3,8). In the present case, the patient reported swelling, paresthesia, and anterior tooth mobility. Jaw expansion and tooth displacement were also noted during clinical examination.

Ameloblastoma shows a marked tendency to involve the mandible (5), which is consistent with our case. However,

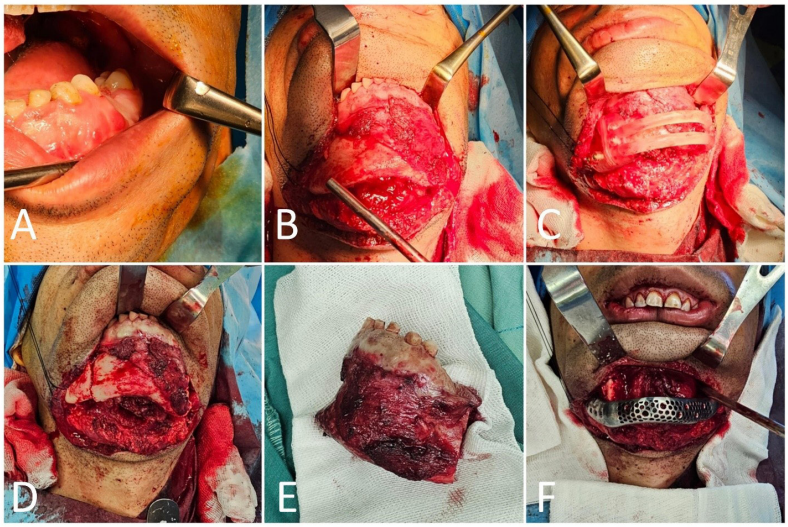

Figure 8. Intraoperative photographs: A) intraoral view prior to surgical resection; B) extraoral approach; C) surgical guide used for resection; D) surgical incisions; E) resected mandibular segment; F) fixation of the patient-specific reconstruction plate.

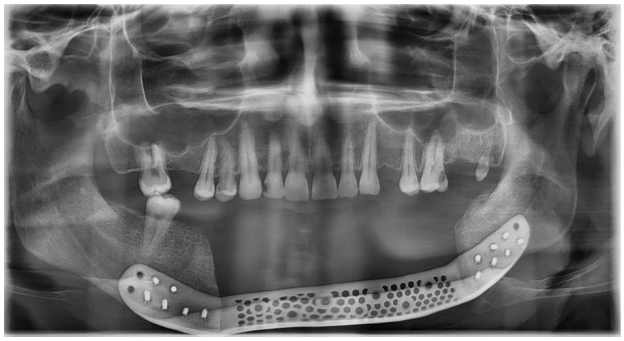

Figure 9. Panoramic Radiograph obtained 3 months postoperatively.

the molar region and ascending ramus are the most commonly affected sites (5,10). In contrast, the lesion in the present case involved the mandibular premolar region as well as the anterior segment, extending across the midline. To our knowledge, this represents one of the rarest presentations of ameloblastoma reported in humans (8).

A thorough understanding of the diverse radiographic characteristics of ameloblastoma is essential for accurate diagnosis (5). In this case, CBCT scan was employed to provide a detailed evaluation of the tumor mass and its extent. Radiographic features of ameloblastoma typically include unilocular or multilocular radiolucencies, with or without cortical thinning and expansion. In the multilocular form, a characteristic "honeycomb" or "soap bubble" appearance may be observed due to the presence of multiple bone compartments created by septa (2,4,6). Root resorption of adjacent teeth is also a common feature (8). In the present case, the lesion exhibited features consistent with ameloblastoma, with the exception of extensive multilocular radiolucencies crossing the midline—an uncommon finding in this tumor. Unlike the desmoplastic variant, other histological subtypes of ameloblastoma generally do not demonstrate significant differences in radiographic features (5). In the present case, the lesion exhibited features consistent with ameloblastoma, with the exception of extensive multilocular radiolucencies crossing the midline—an uncommon finding in this tumor. Unlike the desmoplastic variant, other histological subtypes of ameloblastoma generally do not demonstrate significant differences in radiographic features (5).

Histopathological examination confirmed the diagnosis of acanthomatous ameloblastoma, a rare histological variant characterized by squamous metaplasia and keratinization within the tumor islands.

Treatment options for ameloblastoma range from conservative approaches, such as enucleation and curettage, to radical resection with subsequent reconstruction (4). The recurrence rate has been reported as 16.2% for acanthomatous ameloblastoma (6). Given this risk, radical surgical resection was performed in the present case to minimize the likelihood of recurrence. Patient-specific implants were utilized to stabilize the residual mandibular segments, offering several advantages over conventional reconstruction plates, including a reduced risk of plate exposure and improved facial aesthetics due to their anatomical conformity. Furthermore, these implants facilitate the preservation of adequate space for future bone augmentation and occlusal rehabilitation. Although immediate free microvascular reconstruction is generally recommended, the surgical team elected a delayed reconstructive approach, primarily due to uncertain surgical margins and the potential risk of wound infection. In this case, bone grafting was deferred until recurrence of the lesion could be definitively excluded. Following a one-year surveillance period, definitive mandibular reconstruction will be planned using either a free vascularized bone flap or a particulate iliac crest bone graft to restore mandibular continuity.

Conclusion

In conclusion, this report describes a rare case of acanthomatous ameloblastoma involving both the anterior and posterior regions of the mandible with extension across the midline. The case highlights the diagnostic challenges posed by atypical presentations, the limitations of general pathology in evaluating odontogenic lesions, and the necessity of specialist referral for accurate histopathological diagnosis. Advanced imaging with CBCT and definitive histopathological and immunohistochemical analyses were essential for establishing the diagnosis. Radical surgical resection with patient-specific reconstruction was performed to minimize recurrence and optimize functional and esthetic outcomes.

Declaration of generative artificial intelligence (AI) and AI-assisted technologies in the writing process

The authors utilized ChatGPT to check grammar points and language style in writing. The authors reviewed the text for accuracy and take full responsibility for the final content.

Authors' contribution

Conceptualization: Arash Sarrafzadeh, Parto Nasri, Salar Nasr Esfahani, Maryam Mohebiniya, Soheila Jadidi.
Data curation: Arash Sarrafzadeh, Salar Nasr Esfahani, Maryam. Mohebiniya, Soheila Jadidi.
Investigation: Parto Nasri, Salar Nasr Esfahani.
Project administration: Arash Sarrafzadeh, Maryam Mohebiniya.
Supervision: Arash Sarrafzadeh, Maryam Mohebiniya.
Validation: Parto Nasri, Salar Nasr Esfahani.
Writing–original draft: Arash Sarrafzadeh, Maryam Mohebiniya, Soheila Jadidi.
Writing–review & editing: Salar Nasr Esfahani, Maryam Mohebiniya, Soheila Jadidi.

Conflicts of interest

Patro Nasri serves as the Editor-in-Chief of the journal. However, she was not involved in the peer-review or decision-making process for this manuscript. The other authors declare that they have no competing interests.

Ethical issues

This case report was conducted in accordance with the World

Medical Association Declaration of Helsinki. The patient provided written informed consent for publication as a case report. Ethical issues (including plagiarism, data fabrication, double publication) have been completely observed by the authors.

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