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Case Report

A rare case report of recurrent desmoplastic ameloblastoma in the maxilla

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Abstract

Desmoplastic ameloblastoma is a rare histological variant of ameloblastoma, comprising a small percentage of all cases. It differs from conventional ameloblastoma by its unique histopathology and radiological appearance, typically affecting the anterior maxilla. Recurrence is uncommon but clinically significant due to its locally aggressive nature and tendency for ill-defined borders. The present case describes a patient who experienced recurrence in the maxilla after conservative excision in the same site 25 years ago. The patient now presents with a slowly enlarging, painless swelling in the left maxillary region for the past 12 years. Clinical examination revealed a firm, painless swelling with vestibular obliteration in the left maxilla. Radiographic imaging showed a poorly defined, mixed radiolucent-radiopaque lesion involving the left maxilla with extension toward the nasal fossa. Histopathology confirmed desmoplastic ameloblastoma. This case underscores the importance of early recognition, appropriate imaging, complete surgical excision, and regular long-term follow-up.

Keywords: Desmoplastic ameloblastoma, Recurrent, Odontogenic tumor, Maxilla.

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1. Introduction

Ameloblastomas are benign, locally aggressive, polymorphic neoplasms of proliferating odontogenic epithelial origin, accounting for approximately 1% of all jaw cysts and tumors and 11–59% of odontogenic tumors. Clinically, ameloblastomas are categorized into three variants: Conventional solid or multicystic (≈86%), Unicystic (≈13%), peripheral or extraosseous (<1%).

Histologically, several subtypes have been described, including follicular, plexiform, acanthomatous, granular cell, basal cell, and desmoplastic variants.[1]

Among these, Desmoplastic ameloblastoma (DA) represents a rare subtype, accounting for 4% to 13% of all ameloblastomas.

Desmoplastic ameloblastoma differs markedly from other variants in anatomic location, radiographic pattern, and histologic presentation. While conventional ameloblastomas typically arise in the posterior mandible, DA has a predilection for the anterior maxilla. Clinically, it presents as a slow-growing, painless swelling, and may reach a large size. Radiographically, DA often presents as a mixed radiolucent-radiopaque lesion with ill-defined margins, mimicking fibro-osseous lesions such as ossifying fibroma or fibrous dysplasia.[2]

This variant was first described by Eversole *et al*. in 1984, who characterized it by prominent stromal desmoplasia and the presence of small nests, cords, and strands of odontogenic epithelium embedded in dense collagenous stroma.

Histologically, DA exhibits dense collagenized stroma (desmoplasia) with compressed, irregular islands and strands of odontogenic epithelium. These features may resemble

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odontogenic fibroma, complicating diagnosis.[3] However, the age and sex distribution of DA do not significantly differ from those of conventional ameloblastomas.

2. Case Report

A 60-year-old male reported to the department of oral medicine and radiology with a gradually enlarging, painless swelling in the left maxillary region, present for the past 12 years. He had undergone surgery in the same region 25 years earlier for a similar swelling. Patient give no history of any discharge or numbness. On extraoral examination, facial asymmetry was evident due to a unilateral swelling involving the midface, causing elevation of the nasal floor, lateral displacement of the ala, and loss of the nasolabial fold contour.(**Figure 1**) Intraorally, a dome-shaped swelling extended from the mid-palatal raphe to the buccal vestibule mediolaterally and from the anterior alveolus to the maxillary tuberosity anteroposteriorly.(**Figure 3**) On palpation, the swelling was firm to hard in consistency, with a smooth surface, diffuse margins, and was fixed to the underlying tissues. It was non-compressible, non-tender, and exhibited no local rise in temperature.

A panoramic radiograph revealed a 5.5 × 7.4 cm mixed radiolucent–radiopaque lesion with ill-defined margins involving the left maxilla, crossing the midline and obliterating the left maxillary sinus, along with displacement of the lateral wall of the nasal fossa.(**Figure 3**) CBCT demonstrated an expansile mixed lesion with cortical perforation and involvement of the alveolar ridge, palatal bone, nasal cavity, and maxillary sinus.(**Figure 4**) CT confirmed an osteolytic mass measuring 6.5 × 7.1 × 6.2 cm, with focal cortical breaches and infiltration into adjacent bony structures.(**Figure 5** A, B) Aspiration yielded 2 mL of hemorrhagic fluid, and cytological analysis showed cystic macrophages, inflammatory infiltrates, and necrotic debris, suggestive of a benign cystic lesion.

Based on the patient's history, clinical examination, and radiographic findings, a provisional diagnosis of fibro-osseos lesion was made.

To establish a definitive diagnosis, an incisional biopsy was performed. On microscopic examination it revealed collagen-rich fibrous stroma with scattered islands of odontogenic epithelium and vascular congestion, suggestive of desmoplastic ameloblastoma. The patient underwent partial maxillectomy under general anesthesia. Histopathological examination of the resected specimen confirmed desmoplastic ameloblastoma, showing dense desmoplastic stroma compressing irregular epithelial cords with peripheral palisading, along with areas of hemorrhage, necrosis, and bony involvement. Postoperative healing was uneventful, and a maxillary obturator was provided for rehabilitation. At 5-month follow-up, the patient showed satisfactory recovery without signs of recurrence.

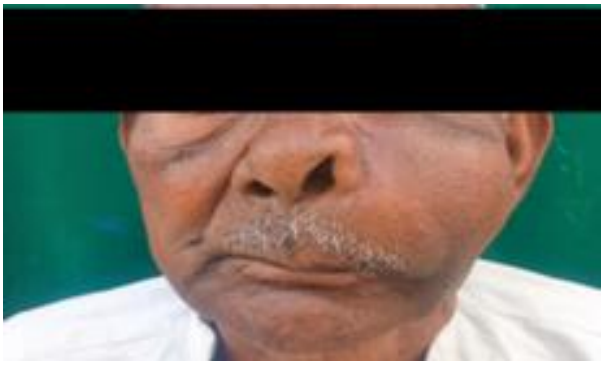

Figure 1: Marked facial asymmetry observed due to unilateral swelling in the midfacial region on extraoral clinical examination

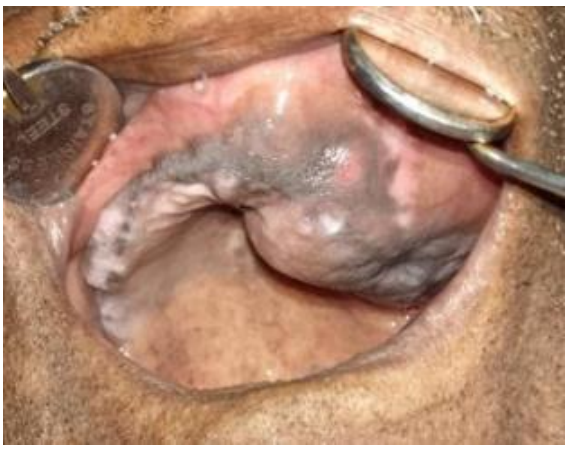

Figure 2: Solitary swelling measuring approximately 6.5 × 6.2 × 4.5 cm, extending from the midline to the left buccal vestibule, causing obliteration

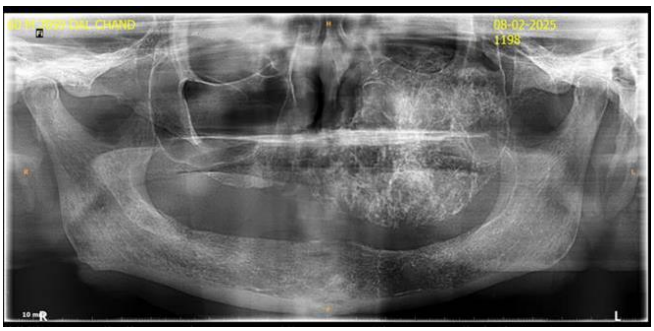

Figure 3: Panoramic radiograph showing a mixed radiopaque–radiolucent lesion involving the left maxilla, extending from the left zygomatic arch and crossing the midline, with ill-defined margins

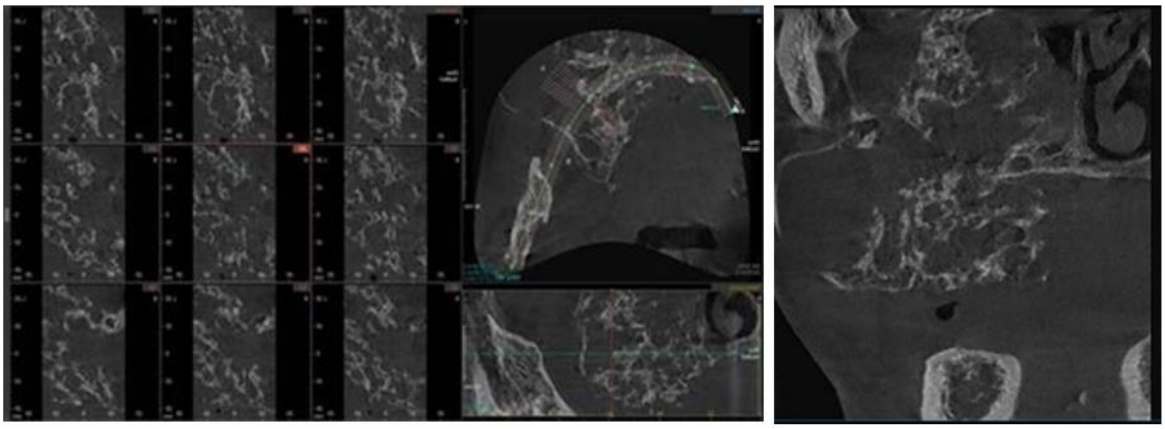

Figure 4: CBCT (coronal and cross-sectional views) showing an expansile lesion with mixed radiopaque–radiolucent internal structure involving the left maxilla and extending inferiorly into the alveolus

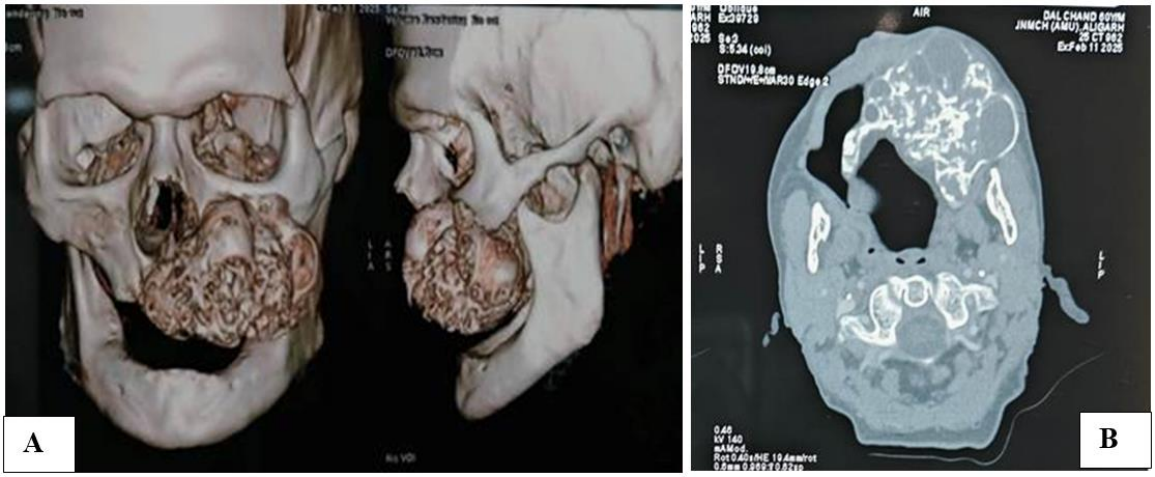

Figure 5: A): 3D volume-rendered CT image showing extensive bony expansion in the left maxilla. **B)**: Axial CT scan image corroborating the findings of extensive bony expansion in the left maxilla

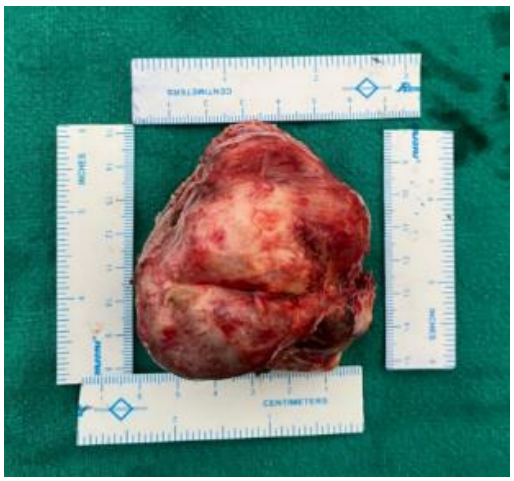

Figure 6: Gross resected tissue specimen submitted for histopathological examination

3. Discussion

In 1984, Eversole first described desmoplastic ameloblastoma (DA), reporting three cases and referring to it as "ameloblastoma with pronounced desmoplasia." Later, in the 2005 WHO classification of odontogenic tumors, DA was recognized as a distinct clinicopathological type of ameloblastoma, alongside the solid/multicystic, extraosseous, and unicystic variants.

The term "hybrid lesions" was introduced by Waldron and El-Mofty to describe cases where DA is found alongside

other types of ameloblastoma, such as the follicular or plexiform patterns.[4,5]

Unlike conventional ameloblastomas that typically affect the mandible, desmoplastic ameloblastomas have a marked predilection for the anterior maxilla in the premolar region of the jaw, often presenting with diffuse swelling and bony expansion without associated pain. In the present case, the lesion presented as a long-standing, painless swelling in the left maxillary region with gradual facial asymmetry and obliteration of adjacent anatomical landmarks, consistent with the slow-growing nature of desmoplastic ameloblastomas.

In a review of 115 cases of DAs, the commonly observed radiographical features were mixed radiolucent-radiopaque (56%), multilocular (49%), and with ill-defined borders (64%).[6]

In this case, the radiographic appearance was typical, showing a mixed radiolucent–radiopaque lesion with ill-defined margins, mimicking fibro-osseous lesions such as ossifying fibroma or fibrous dysplasia, which are common differentials.[7] This underlines the diagnostic challenge posed by desmoplastic ameloblastomas, which often lack the classic soap-bubble or honeycomb radiolucency seen in conventional ameloblastomas.

CBCT and CT imaging further revealed extensive bone expansion, sinus obliteration, palatal bone involvement, and cortical breach, emphasizing the invasive potential of the lesion, despite its benign classification. The aspirated hemorrhagic fluid with cystic macrophages suggested cystic degeneration, which is occasionally reported in desmoplastic ameloblastomas, although it is not a consistent feature.

In this case, both incisional and excisional biopsy, the tumor exhibited the hallmark features of desmoplastic ameloblastoma—dense collagenized stroma (desmoplasia) compressing irregular nests and strands of odontogenic epithelium. These were in consistent with a confirmatory diagnosis of DA that was made by histopathological evaluation. The microscopic features usually include: (1) stromal desmoplasia, in the form of moderately cellular, fibrous connective tissue with abundant collagen, which is the most consistent and distinguishing feature; (2) Islands of different shapes of odontogenic epithelium; (3) peripheral layer of cuboidal cells; and (4) hypercellular central area composed of spindle-shaped or polygonal epithelial cells.[8,9]

Desmoplastic ameloblastoma (DA) demonstrates a locally aggressive behavior and a tendency for recurrence, particularly when managed with conservative treatment approaches. According to Philipsen *et al*., who reviewed over 100 published cases, the recurrence rate of DA is approximately 21.4%, especially when treated with curettage or enucleation, underscoring its infiltrative nature and the importance of aggressive surgical management.[10] Similarly, Pogrel *et a*l. reported a recurrence rate ranging from 15% to 20%, advocating for wide local resection as the treatment of choice to minimize recurrence.[11] Although desmoplastic ameloblastoma generally presents a lower recurrence rate compared to conventional solid or multicystic ameloblastomas—whose recurrence can range from 33% to as high as 90% in mandibular lesions and nearly 100% in maxillary lesions treated by curettage—its potential for extensive local invasion, especially in the maxilla, demands cautious treatment planning.[12] In our case, this was evident from the rapid horizontal and vertical spread of the tumour to involve the zygomatico-maxillary complex and adjacent structures, increasing the chance of recurrence if not treated radically.

Treatment of desmoplastic ameloblastoma generally involves wide local excision or partial maxillectomy, as conservative measures like enucleation or curettage have a high recurrence rate due to the lesion's infiltrative growth pattern. In this case, a partial maxillectomy was performed under general anesthesia, followed by prosthetic rehabilitation with an obturator, restoring function and aesthetics.

4. Conclusion

Desmoplastic ameloblastoma is a rare histologic variant of ameloblastoma that poses significant diagnostic and therapeutic challenges due to its atypical presentation and aggressive local behavior. Its predilection for the anterior maxilla and mixed radiographic appearance can mimic other fibro-osseous lesions, often leading to delayed diagnosis. Early diagnosis, wide surgical excision, and prosthetic rehabilitation remain the cornerstones of effective management. Given the potential for recurrence, long-term clinical and radiological surveillance is crucial. A multidisciplinary approach is essential for accurate diagnosis and optimal outcomes in such rare odontogenic tumors.

5. Source of Funding

None.

6. Conflict of Interest

None.

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