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## INTERNATIONAL JOURNAL OF ADVANCED RESEARCH (IJAR)

Article DOI: 10.21474/IJAR01/18561

DOI URL: <http://dx.doi.org/10.21474/IJAR01/18561>



### RESEARCH ARTICLE

#### SURGICAL MANAGEMENT OF DESMOPLASTIC AMELOBLASTOMA: CONSERVATIVE OR RADICAL APPROACH

Moujoud C., Kissi L., Slimani F., Rifki C. and Ben Yahya I.

#### Manuscript Info

##### Manuscript History

Received: 20 February 2024

Final Accepted: 23 March 2024

Published: April 2024

#### Abstract

Ameloblastoma is a benign odontogenic tumor of epithelial origin. It is locally aggressive with unlimited growth capacity and a high potential for malignant transformation and metastasis. (1) Nowadays, there are two opposing approaches to the surgical management of ameloblastoma. The conservative approach consists of enucleation and/or curettage and the radical approach consists of bone resection with margins of at least 1cm (segmental or not). (2) In this work, we will present a case of desmoplastic ameloblastoma diagnosed in 2014. The patient had been benefiting from regular follow-up. However, a recurrence occurred in 2021. This paper delves into our experiences in managing desmoplastic ameloblastoma, initially employing a conservative approach in 2014 and later transitioning to a radical approach in 2021 in response to the recurrence.

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#### Introduction:-

The ameloblastoma is a benign odontogenic tumor of epithelial origin characterized by its local aggressiveness, unlimited growth capacity, and a high potential for malignant transformation and metastasis. [1][2][14] The management of ameloblastoma is complicated by the significant recurrence rate associated with conservative treatment.[2][5][15]

The World Health Organization (WHO), in its 2023 classification of odontogenic tumors, categorizes ameloblastoma into 5 groups: Ameloblastoma, unicystic; Ameloblastoma, extraosseous; Adenoid ameloblastoma; Metastasizing ameloblastoma, and Ameloblastoma, conventional. [3]The latter, formerly known as solid or multicystic ameloblastoma, includes various subgroups, notably the desmoplastic ameloblastoma [5][16].

This article reports a case of desmoplastic ameloblastoma treated by enucleation in 2014 and by segmental mandibulectomy in 2021 following recurrence.

#### Observation:-

A 50-year-old female patient presented at the Surgical Odontology Department of Ibn Rochd University Hospital in Casablanca in 2014 with a right mandibular swelling that had been developing over 6 months. The patient had no particular medical history. Exobuccal clinical examination revealed a cutaneous swelling under the right lower lip (Fig. 1). No lymphadenopathy was present.

Corresponding Author:- Moujoud Chaimaa

Endobuccal clinical examination showed a firm, non-painful vestibular swelling on palpation in the right mandibular region, extending from tooth 42 to 45 (Fig. 2). Teeth 43 and 45 were mobile. Panoramic radiography showed a poorly defined lytic lesion containing calcified trabeculae extending from the distal aspect of tooth 43 to the distal aspect of tooth 45. The roots of teeth 43 and 45 were displaced by the lesion. No root resorption was observed (Fig. 3). Axial section dental CT scan revealed a multilocular image blowing out the outer cortical with a honeycomb appearance panoramic reconstruction showed the disappearance of the lamina dura at teeth 43 and 45. (Fig. 4). The treatment consisted of enucleation, bone curettage, and extraction of teeth 42, 43, and 45 under local anesthesia. Histological examination showed an ameloblastic proliferation, composed of cellular masses and cords sometimes cystic or crushed within a dense fibrous stroma without atypia or mitoses (Fig. 5). The diagnosis of desmoplastic ameloblastoma was made. Postoperative radiological control at 12 months showed complete bone healing and no recurrence of the lesion. (Fig.6)

In 2021, the patient presented to our consultation with a right mandibular swelling associated with pain without lymphadenopathy, progressively evolving over the course of one year (Figure 7).

Exobuccal examination revealed facial asymmetry due to a low genian swelling on the right side without palpable lymph nodes. The swelling was hard, non-depressible, and painless on palpation.

Endobuccal examination identified an anterior vestibular swelling with posterior extension, accompanied by a bulging of the vestibular cortical bone covered by a mucous membrane with a normal appearance. The swelling was firm and painless to touch. All mandibular teeth were absent, except for 33/37 and 47 (Figure 8).

The panoramic radiograph displayed a multigeodic honeycomb image extending from the location of 34 to the mesial aspect of 47, reaching the basilar margin in a coronal-apical direction. Additionally, a radiolucent image surrounding the roots of 47 was observed (Figure 9).

The 3D reconstruction illustrated the lesion's extension, closely approaching the basilar bone margin. Frontal sections depicted the multigeodic nature of the lesion and revealed the rupture of the internal table in certain areas (Figure 10).



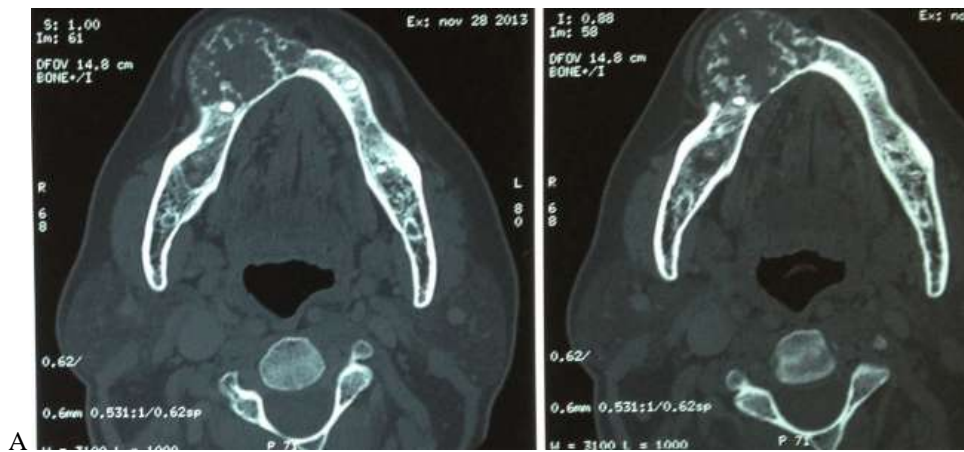
**Figure 1:-** Exo buccal view: right sub-labial swelling.



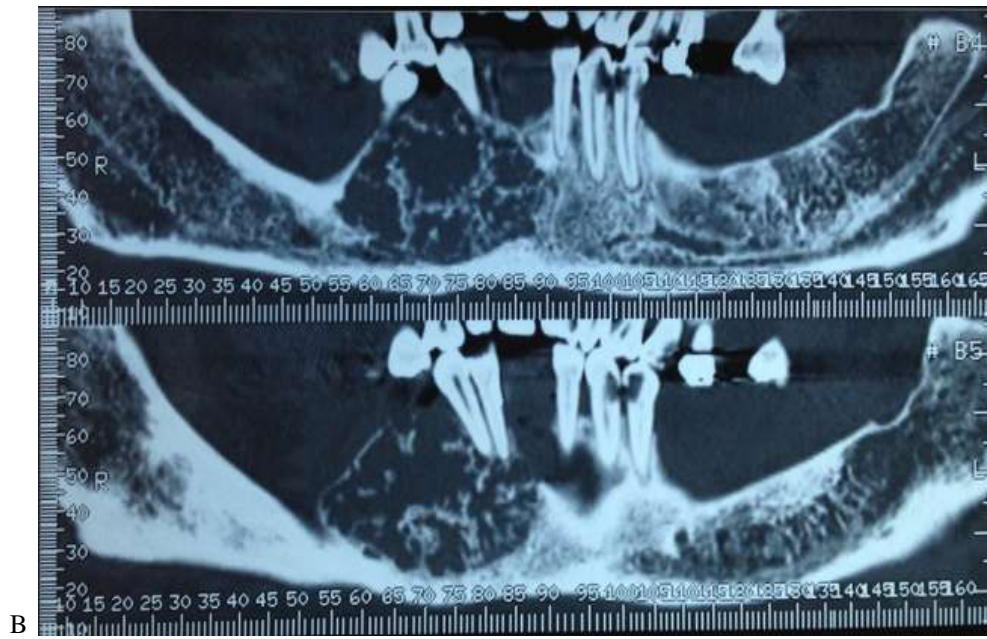
Figure 2:- Endobuccal view: Firm swelling noted in the right mandibular region upon palpation.



Figure 3:- Panoramic radiograph: Indistinct mixed osseous lesion observed between teeth 43 and 45.



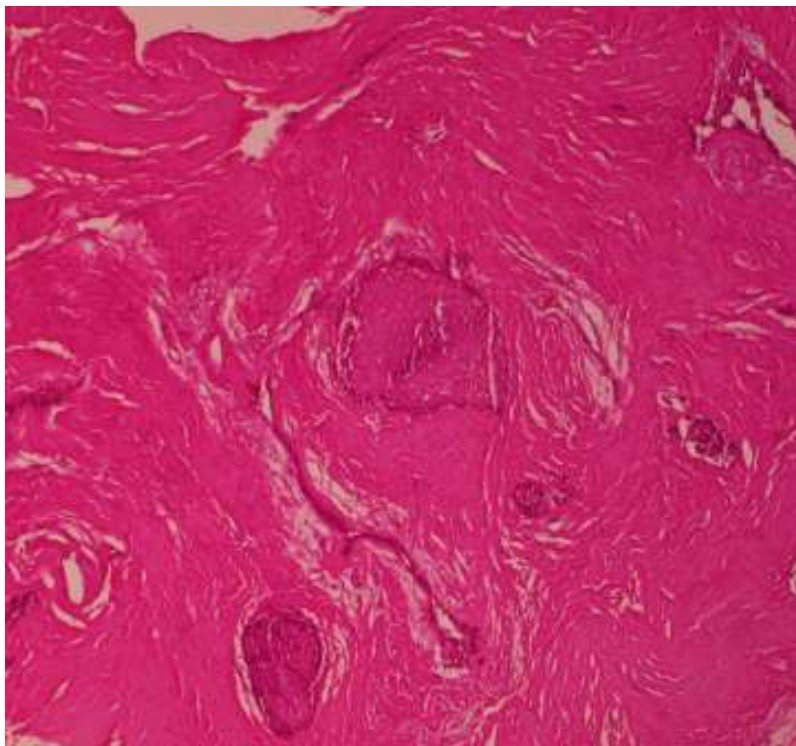




**Figure 4:-**

A: Dental scan in axial sections: Multilocular honeycomb-like lesion.

B: Panoramic reconstructions revealing the disappearance of the lamina dura adjacent to teeth 43 and 45.



**Figure 5:-** Histological examination (H&E staining, 100×): Desmoplastic ameloblastoma.



**Figure 6:-** 12-month postoperative follow-up orthopantomogram: Complete osseous healing.



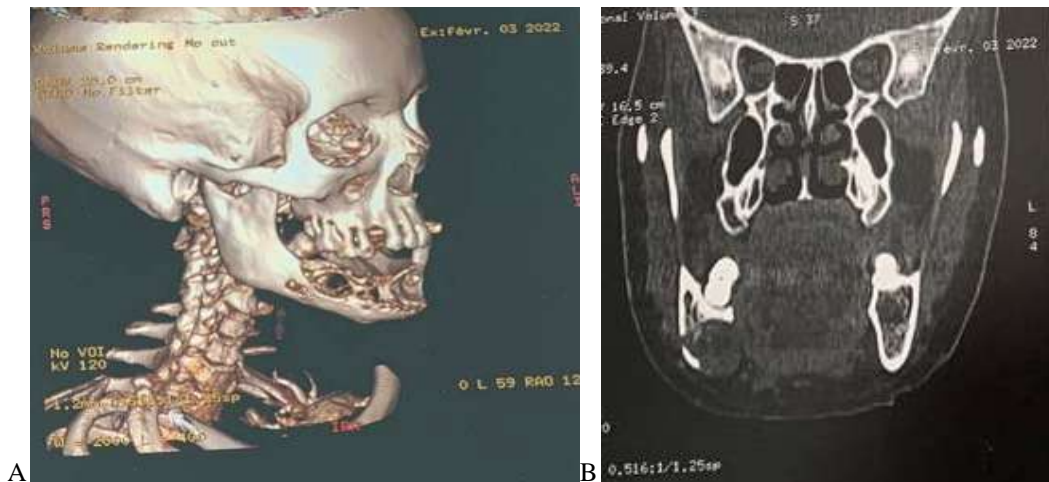
**Figure 7:-** Exobuccal view displaying a low right genian mandibular swelling.



**Figure 8:-** Endobuccal view revealing an anterior right vestibular swelling with cortical bulging.



**Figure 9:-** Panoramic radiograph showing a multi-geodic radioclar image.



**Figure 10:-**

A: The 3D reconstruction showing the extent of the lesion and its intimate relationship with the mandibular basilar bone.

B: frontal slices showing the rupture of the internal table.

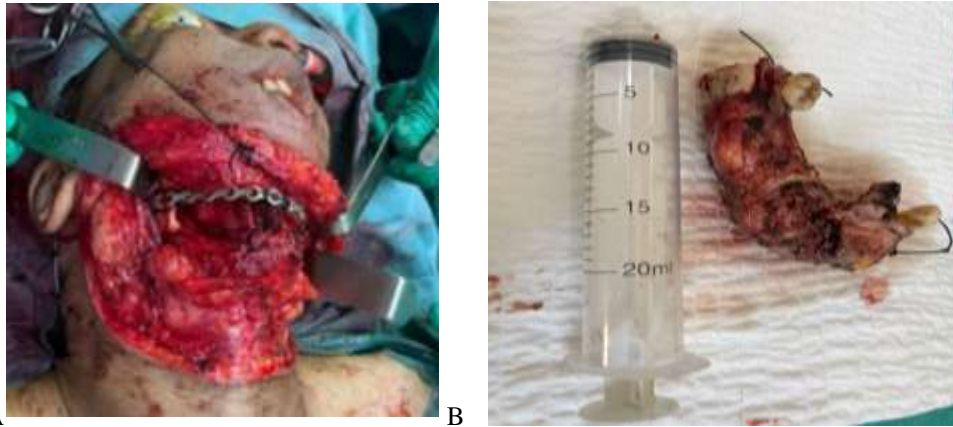
In view of these clinical and radiological elements, we evoked the diagnosis of a recurrent form of desmoplastic ameloblastoma.

The treatment consisted of a segmental mandibulectomy with placement of a titanium plate under general anesthesia in the maxillofacial department on August 20, with sending of the surgical specimen to the analysis laboratory for anatomopathological examination, which was in favor of a desmoplastic ameloblastoma . (Figure 11)

Postoperative radiological control at 12 months showed complete bone healing and no recurrence of the lesion (Figure 12). However, the patient complains of functional repercussions from the radical surgery on his lifestyle. (Figure 13)

Eighteen months later, the patient underwent a skin exposure of the titanium plate, requiring a reoperation for its removal.





**Figure 11:-**

A: Cervical approach for segmental interrupting mandibular surgery under general anesthesia  
B: The surgical specimen



**Figure 12:-** Control radio after 12 months showing complete bone healing.



**Figure 13:-** Frontal View of the patient demonstrating aesthetic defect following Radical Approach.

## Discussion:-

The ameloblastoma is a benign odontogenic tumor arising from epithelial tissue, constituting approximately 1% of maxillary tumors. It commonly presents between the third and fifth decades of life, with a predilection for the mandibular molar region, and an equal incidence among both genders. [1][2][5][14][19]

Desmoplastic ameloblastoma (DA) is an uncommon variant of ameloblastoma, characterized by a remarkably low incidence, reported to range from 0.9% to 12.1% according to existing literature.[17][20]

This tumor often presents in individuals during their fourth and fifth decades, with no gender preference. This subtype typically appears as a smaller lesion in comparison to other types of ameloblastomas. However, when left untreated, it has the potential to increase in size and become highly destructive, requiring extensive excision. The prevalence of DA is more commonly observed in the anterior sections of the jaws, and there is no substantial difference noted between the maxilla and mandible. [17] [19][20]All these data align with the findings presented in this case, as the 50-year-old female patient exhibited a desmoplastic ameloblastoma located in the anterior mandible with a high potential for destructive growth, necessitating extensive excision

Desmoplastic ameloblastoma does not differ clinically from other histological forms of conventional ameloblastoma. It presents mainly as a swelling that is most often painful and slowly evolving [5][17][19][20]. Depending on its evolution and the time of consultation, we find either a swelling only endo-buccal without facial asymmetry, or a facial deformation (as in the reported case) with a swelling that can reach 200x120x130 mm as the case described by Maryame Abou-Elfadl et al[4].Dental mobility and/or displacement may also be present when teeth are involved.[1][5][14]

Our patient exhibited facial asymmetry due to the presence of swelling on the right genian region, accompanied by pain and a bulging of the vestibular cortical bone.

The radiological appearance is not specific. It varies according to the type of ameloblastoma. A 3D examination is necessary to objectify the tumor's relationship in all planes of space. Ameloblastoma most often presents as unilocular or multilocular radiolucent lesions with thinning and expansion of the bone cortices [1][2][5]

In a recent article that reviewed 115 cases of desmoplastic ameloblastomas (DAs), common radiographic characteristics were identified, comprising mixed radiolucent/radiopaque patterns (56%), a multilocular appearance (49%), and ill-defined borders (64%). In the specific case discussed, there was the presence of a multilocular radiolucency with radio-opaque areas noted in the mandibular anterior region. [17][19][20][21]

In this case,histological examination affirmed the diagnosis of Desmoplastic Ameloblastoma. The microscopic features typically encompass: (1) stromal desmoplasia, characterized by moderately cellular fibrous connective tissue with abundant collagen, which is the most consistent and distinguishing feature; (2) islands of odontogenic epithelium with varying shapes; (3) a peripheral layer of cuboidal cells; and (4) a hypercellular central area composed of spindle-shaped or polygonal epithelial cells.[19][20]

The differential diagnosis of ameloblastoma must be made with other non-mineralized odontogenic tumors such as odontogenic myxoma, maxillary cysts and benign non-odontogenic maxillary lesions [5] [7]

Today, there are two opposing approaches to the surgical management of ameloblastoma: The conservative and radical approach. [6]The conservative approach may consist of enucleation and/or curettage, sometimes preceded by marsupialization, for small or moderate tumors with sufficiently thick bone tables. Several studies suggest the use of radiotherapy, cryotherapy, targeted therapy or the Carnoy solution as adjuvant treatment to the conservative approach to increase its chances of success. [10] [11][12]

The radical approach consists of a bone resection with margins of at least 1cm (segmental or not) by:

1. Non-interrupting mandibulectomy by marginal resection:This procedure aims to remove the tumor from the bone by passing at a distance from it, thus in healthy tissue, but without interrupting the mandibular continuity.
2. Segmental Interrupting Mandibulectomy: With this technique, an intermediate segment of the mandibular arch is removed, leaving bone ends on either side of the loss of substance that can be used to support a reconstruction. The integrity of both TMJs is thus preserved. This is the case of our patient



3. Terminal interrupting mandibulectomy: No posterior bone fragment can be substituted to support a reconstruction. The resected mandibular fragment includes at least, by definition, the upper part of the ascending branch with the condylar region. It is accompanied by a disarticulation. A rehabilitation procedure can be considered at the same time. It consists of removing a bone fragment and a muscle flap, generally from the fibula, to reconstitute the resected fraction. [8]

The decision between conservative and radical approaches in ameloblastoma management is challenging and hinges on the tumor's recurrence propensity. Surgeons grapple with whether to adopt a conservative or radical stance, and numerous studies have explored this dilemma.

A 2018 systematic review by Hendra FN et al focused on intraosseous ameloblastoma, showing an 8% recurrence rate with radical treatment compared to 41% for conservative treatment, especially for multicystic ameloblastoma. Another study by Almeida et al, a systematic review and meta-analysis concentrating on multicystic or conventional ameloblastoma, found a 3.15 times higher risk of recurrence with conservative treatment than with radical treatment. Notably, there was no significant difference in recurrence risk between deep marginal resection without mandibular interruption and segmental resection with or without reconstruction. The recommendation was to preserve mandibular continuity as much as possible after bone resection. [5][13]

In our case, due to the young age of the patient and her specific request for management under locoregional anesthesia, our decision in 2014 was to perform curettage exceeding the limits of the tumor to minimize the risk of recurrence. Unfortunately, despite this approach, a recurrence occurred in 2021. Given the extent of the lesion, we referred the patient for management to the maxillofacial service (August 20th), which decided to perform interruptive surgery.

Reconstructing segmental tissue loss is essential, with anterior defects impacting vital prognosis and posterior ones affecting quality of life. Titanium plates remain a straightforward and quick method for mandibular reconstruction. Nonetheless, they entail the risk of exposure, either mucosal or cutaneous, as was observed in the case of our patient. [22]

The recurrence of ameloblastoma, aside from the potential for increased aggressiveness, can result in two significant complications: metastases, often occurring in the lungs, or transformation into an ameloblastic carcinoma. [5]

### **Conclusion:-**

In conclusion, ameloblastoma, as a rare variant, exhibits a polymorphic radiological appearance that can pose diagnostic challenges. The prognosis of this condition depends on various factors, including the patient's age, radiological features, surgical approach, and histological characteristics. Therefore, appropriate management and regular follow-up are crucial for the effective assessment and handling of this complex pathology

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