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Desmoplastic Ameloblastoma: A Case-Series of Three Cases

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1. Abstract

The desmoplastic ameloblastoma is an uncommon subtype of ameloblastoma. Clinical and radiographic presentation differs from other forms of ameloblastoma, and the lesion is sometimes confused with fibro-osseous lesions, making it important for clinicians to review its distinctive clinical and radiographic features. The authors in this article present three cases of desmoplastic ameloblastomas, which were treated at the Augusta University Oral and Maxillofacial Surgery clinic.

2. Introduction

The ameloblastoma is the most common odontogenic neoplasm, superseded in frequency only by odontomas, which are non-neoplastic hamartomatous odontogenic lesions [1,2]. In 2017, the World Health Organization (WHO) simplified the lesion's classification into three categories: ameloblastoma (without a modifier; hereafter referred to as "conventional ameloblastoma"), unicystic ameloblastoma, and extraosseous / peripheral ameloblastoma. Of these three categories, the most common is the conventional ameloblastoma, with histopathologic subtypes including follicular, acanthomatous, plexiform, granular cell, basal cell and desmoplastic [3]. The desmoplastic ameloblastoma (DA) is a relatively rare variant accounting for 4-13% of all ameloblastomas. The lesion presents with notable radiographic and histological deviations from the conventional type, which can render diagnosis and management of DA challenging in some cases. The intent of this article is to document three cases of desmoplastic ameloblastomas, which presented to the Augusta University Oral and Maxillofacial Surgery Clinic, and to review the relevant literature on this rare odontogenic tumor.

3. Case Reports

3.1. Case 1

A 54-year-old African American male was referred to the Augusta University Oral and Maxillofacial Surgery clinic for evaluation of a right mandibular jaw lesion originally diagnosed radiographically as "fibro-osseous lesion." A biopsy submitted from the patient's private practitioner revealed the lesion to be a desmoplastic ameloblastoma. Clinical examination revealed a non-tender, palpable, and expansile bony lesion at the facial surface of the mandible. All other aspects of the physical exam were within normal limits. A maxillofacial contrast CT was ordered (Figure 1a, 1b) which confirmed the presence of a mixed radiolucent / radiopaque lesion of the right mandibular angle approximately 26.4 mm anteroposterior x 12.8 mm craniocaudal x 13.3 mm transverse, with associated erosion of the lateral buccal cortex of the mandibular alveolus. Microscopic evaluation of the resected mandible provided confirmation of negative resection margins as well as confirming the diagnosis of DA. The histopathologic sections demonstrated attenuated follicle-shaped neoplastic islands and thin cords of odontogenic epithelium with deeply-chromatic nuclei, focally surrounded by columnar, ameloblast-like cells exhibiting nuclear reverse polarization. These neoplastic epithelial islands and cords are embedded in a markedly hyalinized eosinophilic acellular stroma interspersed with dense cellular fibroblastic connective tissue (Figure 2a, 2b). The patient underwent a right partial mandibulectomy and placement of a reconstruction plate (Figure 3). Eight months later, mandibular reconstruction was performed using a non-vascularized bone graft from the patient's left anterior iliac crest (Figure 4). The patient had an unremarkable post-operative course.

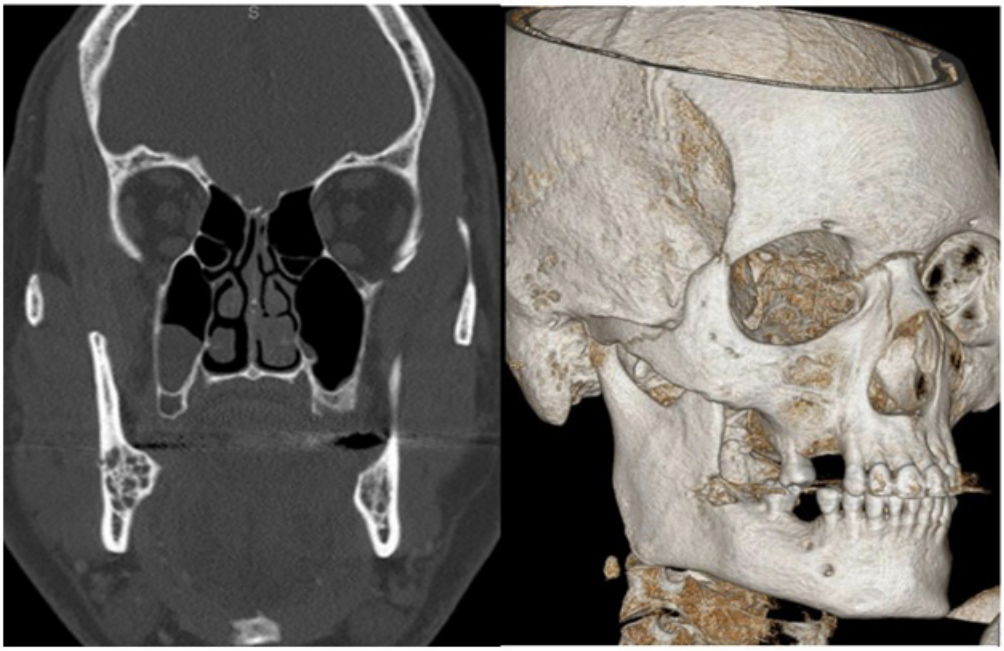


Figure 1a **Figure 1b**

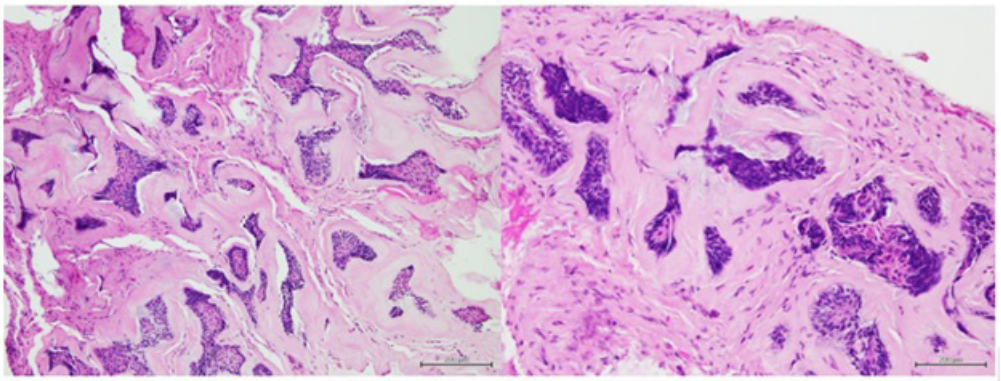


Figure 2a **Figure 2b**

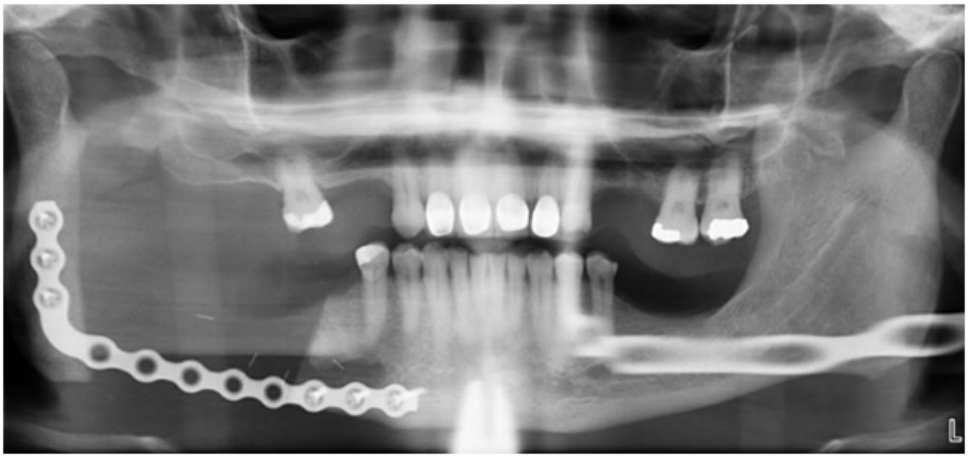


Figure 3

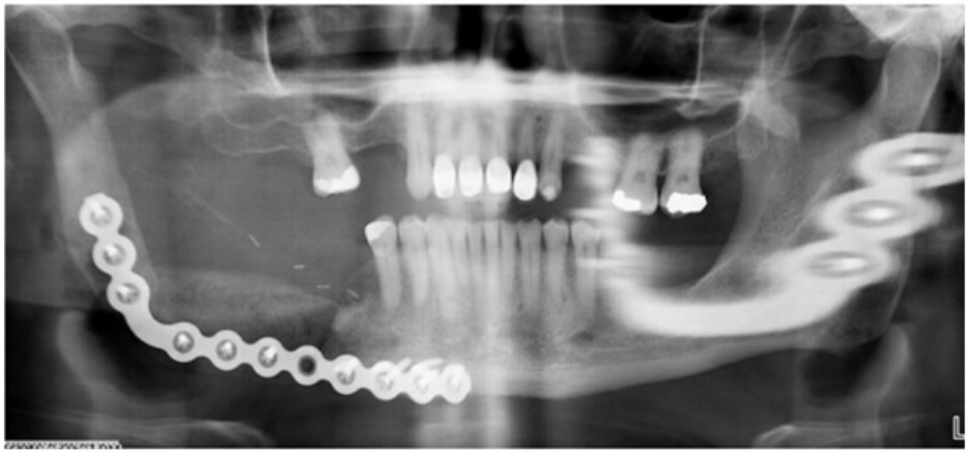


Figure 4

3.2. Case 2

A 57-year-old Caucasian female presented to the Augusta University Oral and Maxillofacial clinic for evaluation of an asymptomatic bony growth lingual to teeth #10 and #11. The patient had a history of treatment in the area, with a similar lesion being removed approximately 15 years prior by a private practitioner. The patient was not aware of what the final diagnosis of that lesion was. A new panoramic radiograph revealed what was assumed to be a recurrence of the lesion (Figure 5). Physical exam showed a 5 mm x 5 mm firm, raised lesion on the palatal mucosa lingual to teeth #10 and 11, with no sinus tracts and appreciable bruit. Periapical radiograph and cone beam computed tomography (CBCT) exams of the region showed a mixed radiopaue-radiolucent 10 mm x 5mm x 5mm lesion separate from the nasopalatine foramina with expansion of the facial and palatal cortical plates (Figure 6, 7). Differential diagnosis included lateral periodontal cyst, calcifying epithelial odontogenic tumor, and adenomatoid odontogenic tumor. A biopsy of the lesion was obtained via enucleation. Histologic examination of the submitted specimen revealed a benign odontogenic infiltrative epithelial neoplasm, characterized by thin attenuated cords and small islands with ameloblastic differentiation embedded in a desmoplastic connective tissue stroma (Figure 8-9). A microscopic diagnosis of DA was made. At a subsequent appointment, an *en bloc* resection of the lesion was recommended. However, the patient has not followed-up since that time, and no definitive treatment has been rendered.

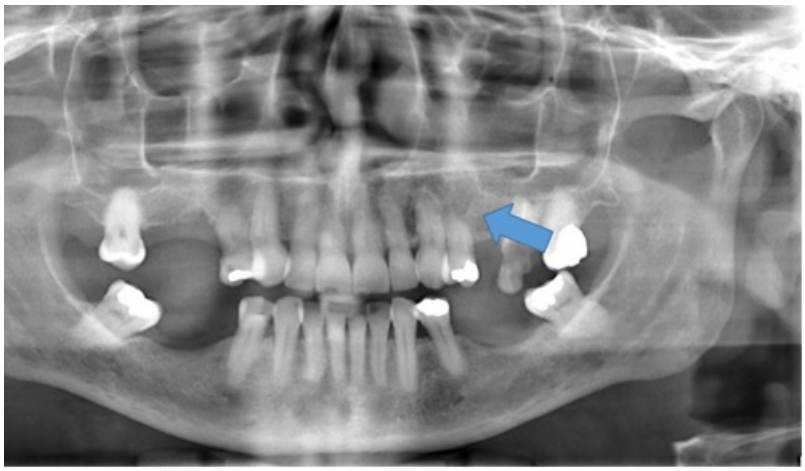


Figure 5

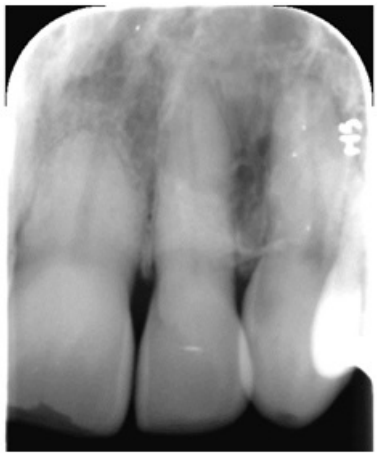


Figure 6

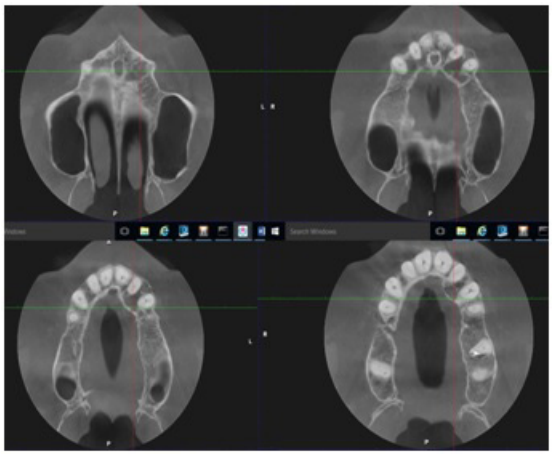


Figure 7

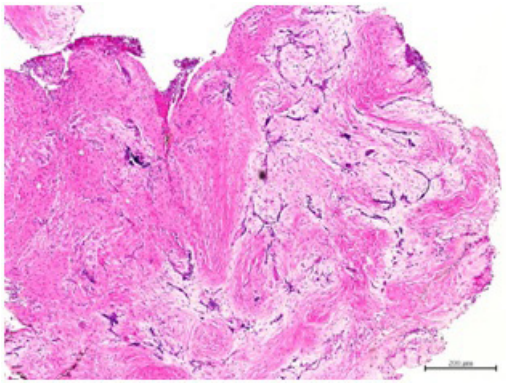


Figure 8

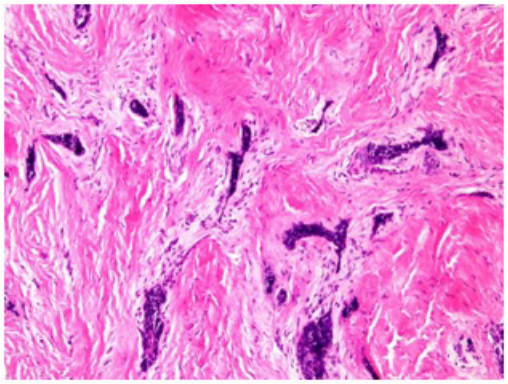


Figure 9

3.3. Case 3

A 37-year-old African American male presented to the Augusta University Oral and Maxillofacial Surgery clinic with a referral from an outside general dentist to evaluate a "cyst" in the left maxilla. The patient reported swelling in the area, which was first noted two years prior. Routine X-ray films and CT images revealed mixed radiolucent-opaque lesion extending from #11-#15 (Figures 10 and 11). The lesion measured approximately 20 mm x 15 mm x 10 mm in the left maxillary quadrant superior to teeth #11-#15 with displacement of the adjacent tooth roots. Clinical examination revealed palatal expansion around sites #13 and #14, surrounding erythema, and Miller 1 mobility of teeth #11-15. The lesion had been biopsied about one month previously by an outside provider. Histologic examination of the submitted lesion revealed a proliferative epithelial odontogenic neoplasm embedded in desmoplastic stroma with interspersed bone spicules. A diagnosis of DA was made.

The patient underwent a left hemimaxillectomy (Figure 12), and the specimen was submitted for histopathologic examination. Histologic examination of the resected specimen confirmed DA.

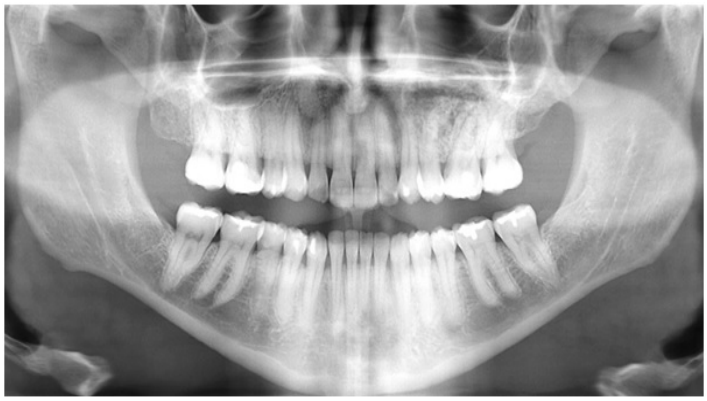
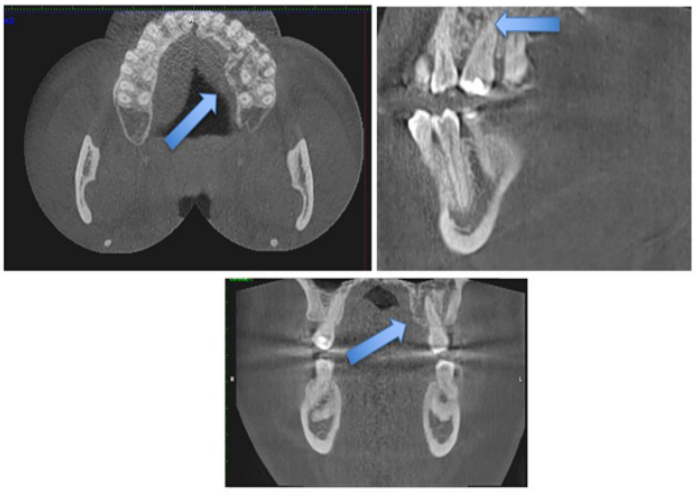


Figure 10



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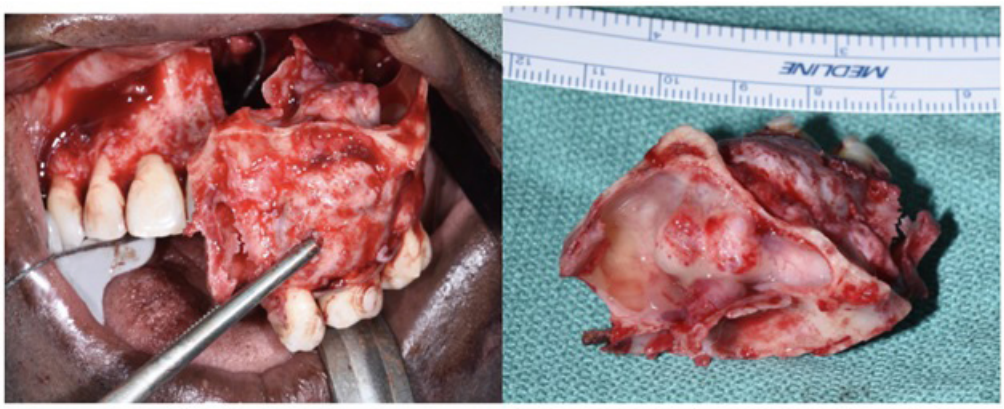

Figure 12

4. Discussion

Ameloblastoma is the most common odontogenic neoplasm. It arises from rests of the odontogenic epithelium, from the enamel organ, from the epithelial lining of odontogenic cysts, or from the basal cells of gingival mucosal epithelium. They are benign neoplasms, but are locally aggressive and infiltrative, and they frequently cause cortical thinning and expansion of the involved region of the jaws along with root resorption of adjacent teeth. Despite this, patients with ameloblastoma are typically asymptomatic even in cases when grotesque growth and expansion occurs. Radiographically, ameloblastomas most often present with an expansile, multilocular, "soap bubble" appearance. Less commonly, they are unilocular, and rarely present as mixed radiolucent and radiopaque lesions. Histopathologic patterns of conventional ameloblastoma include the follicular, acanthomatous, plexiform, granular cell, basal cell and desmoplastic [3]. The subcategory "desmoplastic ameloblastoma" has only been recognized relatively recently, with Eversole being the first to give a detailed description of what he termed "ameloblastoma with pronounced desmoplasia" in 1984 [4]. The World Health Organization initially included DA as its own clinicopathologic entity in 2005, but has since reclassified it as a histological subtype of conventional ameloblastoma owing to its similar course and management [2]. Still, DA presents with clinical, radiographic, and histological features distinct from conventional ameloblastoma. As of 2019, only 244 cases of non-hybrid DA have been reported in the literature [5]. Nearly half of those cases (45.9%) presented in the maxilla, which contrasts with conventional ameloblastoma, which has a marked predilection for the mandible (80%) [6]. Two of the three cases presented in this article were located in the maxilla, highlighting this contrast. The tendency of DA to occur in the anterior maxilla may result in a higher frequency of anatomy-related complications, such as expansion into the maxillary sinus or nasal cavity with resultant unimpeded, rapid growth. Furthermore, the thin cortical bone of the maxilla and the surgical caution necessitated by proximity to important structures may contribute to the higher rate of recurrence for maxillary DA [5], as seen in the recurrence of Case 2. Demographically, DA appear at about equal rates in men and women, most commonly between ages 30-60. Patients commonly report painless swelling, and tooth displacement, observed in about 90% of cases. However, root resorption is only seen in about 1/3 of cases [7]. Radiographically, DA present as one of three types: mixed radiolucent and radiopaque (osteofibrotic type), completely radiolucent, and radiolucent/radiopaque with a large radiolucent change (compound type). Most cases of DA are of the osteofibrotic type. This appearance is presumed to be due to osteoplasia and infiltration of the tumor into the bone trabeculae [7]. On panoramic radiographs, DA may appear similar to fibro-osseous lesions, which can lead to inaccurate diagnoses such as the initial diagnosis of "mixed fibro-osseous lesion" in Case 1. The use of CBCT can be valuable in differentiating DA from other forms of ameloblastomas, showing a honeycomb-like appearance due to the presence of coarse trabecular septa, whereas other forms of ameloblastoma manifest as unilocular or multilocular radiolucent lesions. Histological evaluation is critical in the definitive diagnosis of DA. Small islands and cords of odontogenic epithelium in densely collagenized stroma is the most commonly described pattern [8]. Inconspicuous peripheral columnar cells can be located around the epithelial islands. However, the peripheral layer is typically cuboidal and may present as hyperchromatic. The central regions of the islands are typically composed of spindle-shaped or squamoid epithelial cells. Osteoplasia is also common. The follicular palisading often seen in conventional ameloblastomas is notably absent [9].

5. Conclusion

The cases presented in this article highlight the many key features that differentiate DA from other forms of ameloblastoma. Because DA often resemble fibro-osseous lesions radiographically, the clinician must have a thorough understanding of these features so that proper differential diagnoses are considered. Furthermore, clinical differences between DA and other forms of ameloblastomas are necessary for the clinician to understand, including factors such as location of occurrence, rates of recurrence, and the possibility of invasion into vital structures. Because of these features, resection

is recommended in cases of DA, which results in the lowest rate of recurrence [5].

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