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Desmoplastic Ameloblastoma of Anterior Mandible: A Case Report

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Abstract

Background: Desmoplastic ameloblastoma (DA) is a rare histologic variant of ameloblastoma, accounting for approximately 4 to 13% of ameloblastomas. It is uncommon, aggressive in nature, and there are high chances of misdiagnosing it as a fibro-osseous lesion. Clinical and radiographical features are similar to fibro-osseous lesions of jaw.

Case presentation: A 20 year old male reported to the Out Patient clinic of our department with a painless swelling in the anterior mandible. A mixed lesion with multilocular appearance was evident on occlusal, panoramic radiograph as well as Computed Tomography scan. An incisional biopsy confirmed it to be a case of desmoplastic ameloblastoma.

Conclusions: The present case deserves emphasis because the clinical and radiographic features of desmoplastic ameloblastoma are similar to fibro-osseous lesions and definitive diagnosis should always be based on the

histopathologic findings. Treatment also varies for fibro-osseous lesion and desmoplastic ameloblastoma.

Keywords: Desmoplastic ameloblastoma, histopathology, mixed lesion, odontogenic tumor.

Introduction

Odontogenic tumors are lesions derived from epithelial or ectomesenchymal tissues or both. They range from hamartomatous or non-neoplastic tissue proliferation to malignant neoplasms with metastatic potential. In humans, tumors of the odontogenic tissues are comparatively rare, comprising about 1% of all oral and maxillofacial biopsy specimens [1]

Ameloblastoma is the second most common tumor, only next to odontoma. The tumor is considered benign despite of its locally invasive nature. The follicular and plexiform varieties of ameloblastoma are most common, followed by the acanthomatous and granular cell types. Less frequent cellular variants of ameloblastoma are desmoplastic ameloblastoma, clear cell ameloblastoma, basal cell ameloblastoma and keratoameloblastoma.

Desmoplastic ameloblastoma (DA) is rare; accounting for approximately 4 to 13% of ameloblastomas. It was first described by Eversole *et al*. in 1984 as a new type of ameloblastoma. World Health Organization (WHO) 2005 histological classification of odontogenic tumor categorizes DA as a separate entity and defines it as a variant of ameloblastoma with specific clinical, imaging, and histological features. Desmoplastic ameloblastoma differs from other variants of ameloblastoma in that it is more commonly seen in the anterior region of jaw and its mixed radiolucent radiopaque appearance is often more representative of a fibro-osseous lesion [3].

In this case report, we present a case of DA in a 20-year-old male patient based on typicalclinical, radiological, and histopathological features.

Case Report

A 20 year old male patient reported with a chief complaint of painless swelling in the lower front tooth region for 6 months. On clinical examination, extra orally diffuse firm mild swelling was noted. [Figure 1].Intraoral examination revealed a solitary swelling measuring approximately 3 × 2 cm, extending from the distal aspect of 33, crossing the midline to the mesial aspect of 44 region and from the marginal gingiva to the alveolar mucosa [Figure 2]. Mucosa over the swelling was normal, with, no sinus openings or discharge. On palpation, it was hard in consistency and non tender. No fluid was evident on fine needle aspiration. On radiographic examination, mandibular cross sectional and topographic view showed an expansile multilocular lesion in the mandibular symphysis –parasymphysis region with labial cortical plate expansion and displacement of 43 labially. [Figure 3,4].On further radiographic examination, panoramic radiograph revealed a periapical radiolucent–radiopaque lesion in relation to 32 to 44[Figure 5]. Computed tomography showed ill defined solid expansile multiloculated lesion with honey combing appearance invoving the bilateral parasymphysis, symphysis and anterior part of body of mandible on right side.The root of 43 was pushed labially and seen within the lesion. Expanded labial cortical plate showed erosion with adjacent soft tissue thickening [Figure 6, 7a, 7b, 7c]. Considering the clinical features and mixed radiolucency and opacities, a provisional diagnosis of fibro osseous lesion was made. Incisional biopsy was done under local anesthesia and tissue was sent for histopathological examination. Hematoxylin and eosin stained sections showed connective tissue with collagen fiber bundles, few areas of hyalinization and thin strands and cords of odontogenic epithelial islands containing central cuboidal cells and peripheral flattened epithelial cells [Figure 8].

Correlating with clinical and radiological features, a histopathological diagnosis of DA with respect to anterior mandible was given and en bloc resection was done with respect to mandibular left canine to right premolar region.

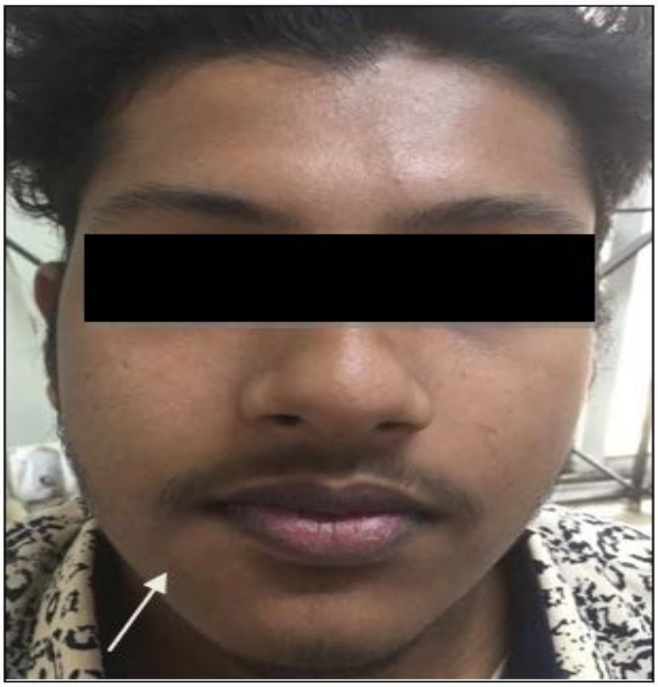

Figure 1: Diffuse mild swelling below the right commissure of lip on extra oral examination

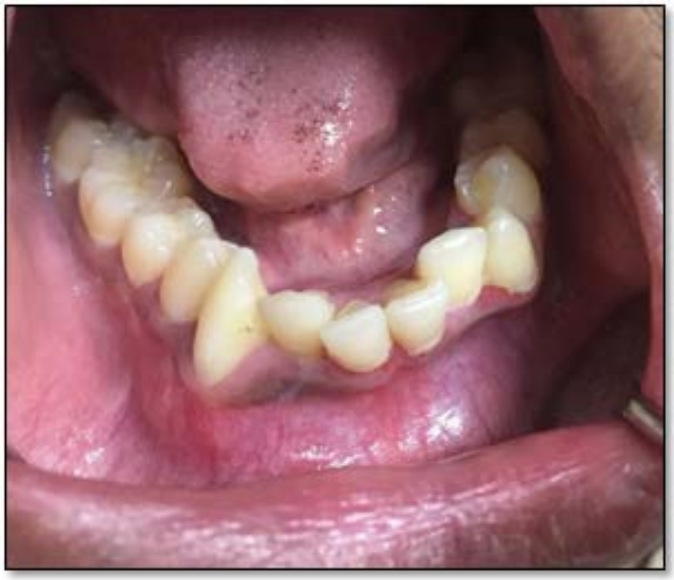

Figure 2: Solitary swelling measuring approximately 3 × 2 cm, extending from the distal of 33 to mesial of 44 and from the marginal gingiva to the alveolar mucosa

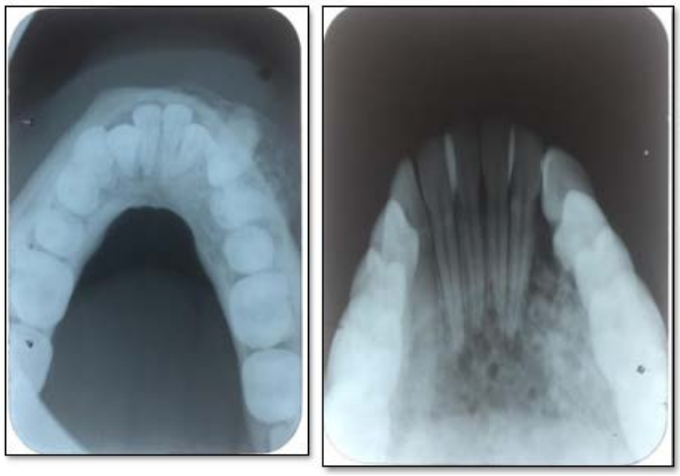

Figure 3, 4: Mixed radiolucent radiopaque appearance with labial cortical expansion and displacement of 43 labially

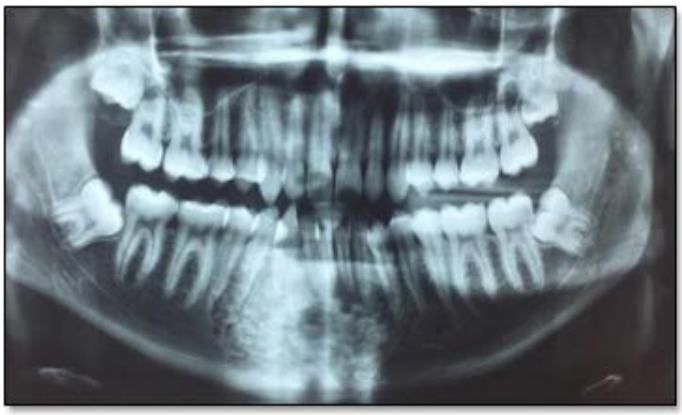

Figure 5: Mixed radiolucent radiopaque appearance involving mandibular alveolus andbasal bone from mesial aspect of 34 to mesial aspect of 45

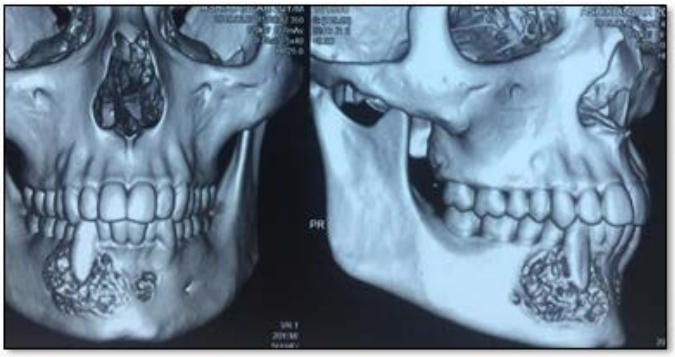

Figure 6: CT 3D View

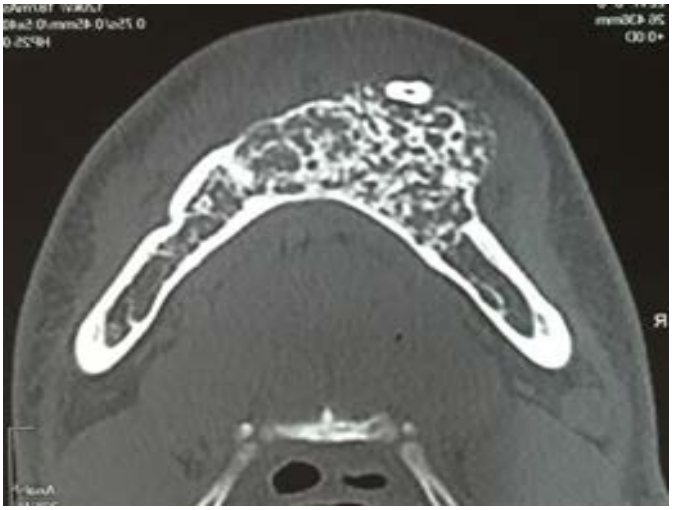

Figure 7a: CT axial section

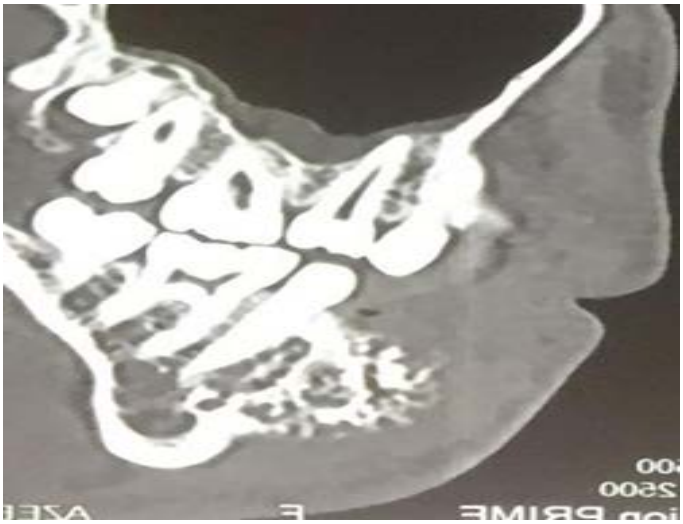

Figure 7 b: CT Sagittal section

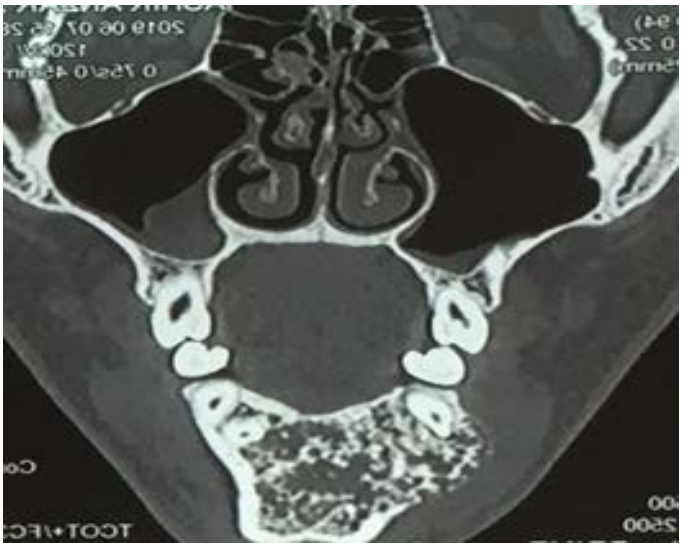

Figure 7 C: CT coronal section

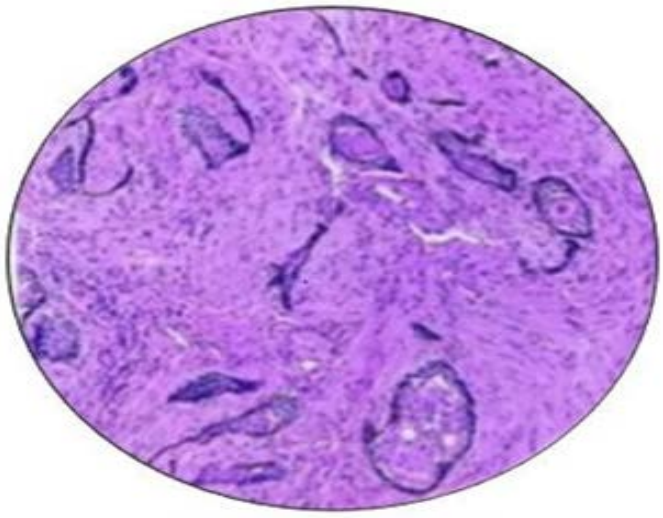

Figure 8: Photomicrograph showing areas of hyalinization and thin strands and cords of odontogenic epithelial islands containing central cuboidal cells and peripheral flattenedepithelial cells

Discussion

Eversole in 1984 first established the report on DA to the English literature and called it an 'ameloblastoma with pronounced desmoplasia'. In the WHO's Histopathological Typing of Odontogenic Tumors 2005, DA is included as a separate clinicopathological entity and classified ameloblastoma into four types as solid/multicystic, extraosseous, desmoplastic, and unicystic.

The term "hybrid lesions" was introduced by Waldron and ElMofty reporting a condition in which DA was present close to follicular or plexiform ameloblastoma and Wakoh *et al*. presented a case of a patient demonstrating follicular type ameloblastoma with desmoplasia, in whom radiological findings suggested the coexistence of a fibro osseous lesion with a solitary cystic lesion and proposed it to be hybrid follicular/DA[4].

The DA is a rare variant of ameloblastoma, which is characterized by marked stromal desmoplasia. This tumor is most commonly found in the fourth and the fifth decades of life, with no sex predilection[5] .Usually,

DA is smaller than other types of ameloblastoma, but if neglected, it can be very extensive and destructive, requiring wide excision. More frequently, it occurs in the anterior part of the jaws, and there is no difference in prevalence between the maxilla and the mandible. The main clinical symptom is apainless swelling with buccal expansion of the mass and teeth displacement [6].

In a recent article, Effiom and Odukoya reported that multilocular radiolucency is the predominant radiographic presentation of the desmoplastic variant of the DA. In contrast,the variant with osteoplasia presented as a mixed radiolucent -radiopaque appearance, thereby mimicking a fibro-osseous lesion.[7]

Histopathologically, irregular odontogenic islands with a stretched-out 'kite-tail' appearance were seen in a dense desmoplastic stroma.The peripheral layer of the epithelial islands was made up of flattened cells and the inner core was made up of spindle-shaped and, in some instances, squamous-shaped cells[8]

In a review of 169 cases of DAs, the commonly observed radiographical features were mixed radiolucent/radiopaque (56%), multilocular (49%), and with ill defined borders (64%).[9] In our case, multilocular radiolucency with radioopaque areas was observed in the mandibular anterior region[9]

The radiological and histopathological findings of poor encapsulation and ill-defined borders suggestive of its infiltrative nature which warrants in depth analysis and a long term follow-up. With potential for recurrences, a complete resection is recommended.

Conclusion

Ameloblastomas may thus be common in the anterior and posterior jaw and the tumors inthe anterior jaw may mature early, explaining the unique site predilection, mixed radiolucent appearance, and histologic presentation of DAs. The clinical and radiographic features of DA are similar to fibro osseous lesions, but definitive diagnosis should always be based on the histopathologic findings. Treatment also varies for fibro osseous lesion and DA. Histopathological diagnosis after incisional biopsy gives a proper channel for the treatment plan.Because of its aggressive nature, a complete resection and long term follow up is required to check any recurrence and even prognosis.

Declaration of patient consent**:** Written informed consent was obtained from the patient for publication of this case reportand any accompanying images.

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