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Case Report

Desmoplasia and Osteoplasia in a Hybrid Ameloblastoma: A Case Report

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Abstract

Desmoplasia in ameloblastoma is comparatively a newer histopathologic variant of ameloblastoma. This variant is characterized by unusual histopathological features and marked stromal desmoplasia. The occurrence of osteoplasia is not frequently seen in ameloblastomas. Our present case is characterized by stromal desmoplasia and osteoplastic changes along with classical follicular, acanthomatous and basaloid differentiation in a tumour arising in a 28 year old female in the anterior region of the mandible.

Keywords: Ameloblastoma; Benign Odontogenic; Desmoplasia; Hybrid Tumour; Osteoplasia; Tumour.

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Introduction

Ameloblastomas are epithelial odontogenic tumors which have been extensively reviewed over years since 1934.[1] Numerous cases have been presented running into thousands of case reports. These throw light on the variety of histological variants and patterns seen in ameloblastoma.[1] They also reflect different behavioral patterns and aggressiveness of the tumor. Follicular and plexiform are the main histological variants. Cytomorphogenic subtypes of the above two main patterns are basaloid, acanthomatous, granular cell, unicystic and more recently, clear cell, papilliferous, kerato-variant and desmoplastic ameloblastoma (DA).[2]

Though it is believed that variation in pattern do not bear any significance in behavior or prognosis, only the unicystic ameloblastoma is said to be less aggressive clinically. Desmoplastic ameloblastoma is a variant of ameloblastoma recognized a few decades ago with few published cases. This variant shows unusual clinical and radiographic findings in comparison to the other variants of ameloblastoma.[3,4] Hence it is suggested that it probably makes up a clinicopathological entity worthy of separate consideration.[3] Interestingly, a possible transitional form of desmoplastic ameloblastoma, showing microscopic areas of desmoplasia with typical areas of classic follicular or plexiform ameloblastoma, has been described as a “Hybrid Lesion”.[4]

The present article reports a case of desmoplastic ameloblastoma with features of stromal desmoplasia with areas of follicular pattern, acanthomatous and basaloid differentiation of conventional ameloblastoma along with predominant osteoplasia.

Case Report

A 28-year-old female patient reported with a slow growing & painless swelling in the left anterior region of the lower jaw since 26 months. A small extra oral swelling was seen on the left cheek near the angle of the mouth below the ala-tragal line measuring approximately 2 cm in diameter. Intra orally, a bony hard swelling, measuring about 2.0 - 2.5cm in dimension, was seen covered with normal appearing mucosa in the region of teeth #33 and #34, was well demarcated and obliterated the labial vestibular region (Fig 1a). Radiographically, the lesion showed a ground glass appearance in the region of teeth #33, #34, and #35 with small areas of radio-opacity and surrounded by ill-defined margins (Fig 1b). Root resorption was not evident in the affected area. The buccal plate expanded into a bulge (Fig 1c).

A provisional diagnosis based on clinical and radiographic findings was made in favor of a benign fibro–osseous lesion. Surgical resection was done under general anesthesia. The lesional mass was ill demarcated and removed out of the surrounding bone with a border of normal bone. Patient was recalled for regular follow-up and no evidence of recurrence was seen in 26 months. On gross examination, the enucleated lesion was well circumscribed,

and on sectioning the firm mass, it showed a gritty surface. The microscopic examination revealed mature connective tissue stroma with marked desmoplasia in many areas. The tumor epithelial cells were seen and were arranged in small to large follicles in an attempt to be compressed. Most of the tumor islands and follicles were laid in a dense collagenized stroma, hence squeezing effect was seen and follicles appeared to be shrinking at the peripheries (Fig 2a). Typical features of follicular ameloblastoma, with peripheral, tall, columnar cells containing palisading nuclei, resembling ameloblasts with a central core of stellate reticulum-like cells, were observed (Fig 2b). Some follicles exhibited squamous metaplasia and basaloid differentiation. In many areas new bone formation was prominently seen with osteocytes within the matrix and a distinct osteoblastic rimming (Fig 2c). So the final diagnosis of Hybrid Ameloblastoma with desmoplasia and osteoplasia was given.

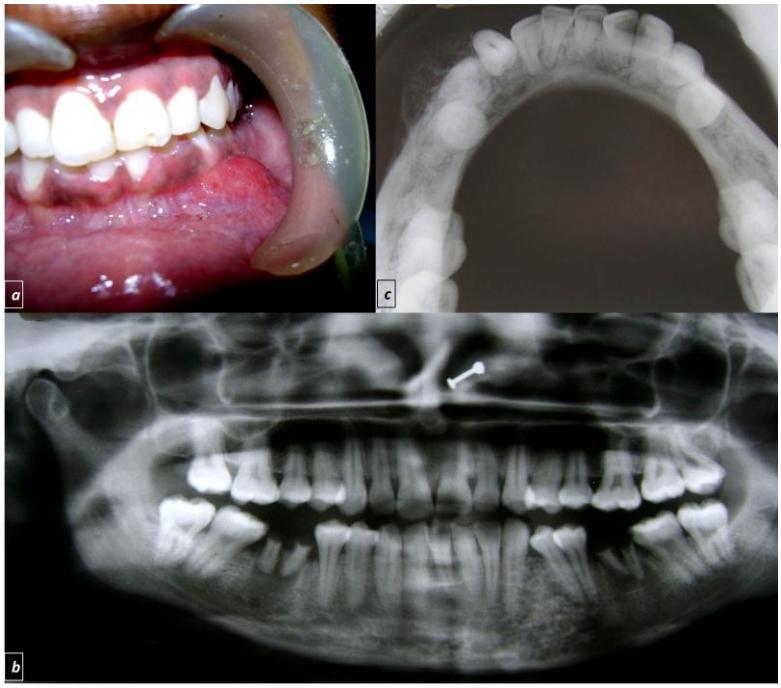

Figure 1: The Clinical swelling of the mandibular left alveolar process obliterating the buccal vestibule extending from canine to second premolar (a). The orthopantamograph shows mottled or bone rarefaction with respect to 33, 34, and 35 region (b) and occlusal radiograph showing mixed radiopaque/radiolucent lesion on the labial aspect of mandible extending from canine to second premolar (c).

Discussion

The mention of pronounced desmoplasia in ameloblastoma drew attention from the initial reports of Takigawa et al.[5] and Uji et al.[6] Eversole described it in detail as an unusual variant of ameloblastoma[7], and later called this variant as desmoplastic ameloblastoma. Hybrid lesions have microscopic features of the desmoplastic variant of ameloblastoma along with areas of classic plexiform, follicular or acanthomatous ameloblastoma.[2] On the basis of the (hybrid) cases published so far the relationship between the desmoplastic ameloblastoma and the hybrid lesion cannot yet be determined.[4] The findings which are common to our present case and previously reported cases are:[8]

- The tumor was seen in a 28-year-old female.
- It was observed in the anterior portion of the jaw.
- The tumor originated in the central area of the bone.
- The tumor was a solid ameloblastoma.
- Histopathology showed typical ameloblastic follicles and follicles with areas of acanthomatous and basaloid differentiation.
- Predominant desmoplasia and osteoplasia was seen in many areas throughout the tumor.

Waldron and El-Mofty undertook an extensive histopathological study of 116 ameloblastomas and their study showed that in desmoplastic variant, the anterior portion of maxilla is more frequently involved.[8] In mandible, it is generally seen in premolar /

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anterior region. In the present case, the tooth bearing area of the bone in the anterior region of the mandible was involved. Most of the cases reported till now were in women and are intra–osseous ameloblastomas.[9] In our case we confer with all the above mentioned features.

Typical radiographic appearance in desmoplastic ameloblastoma resembles a fibro-osseous lesion. In our present case, we observed a localized radiolucency with an indistinct border. The lesional area showed hazy areas of radioopacity. The buccal plate was expanded abruptly but has a smooth border, leading to a probable diagnosis of fibro-osseous lesion rather than an ameloblastoma.

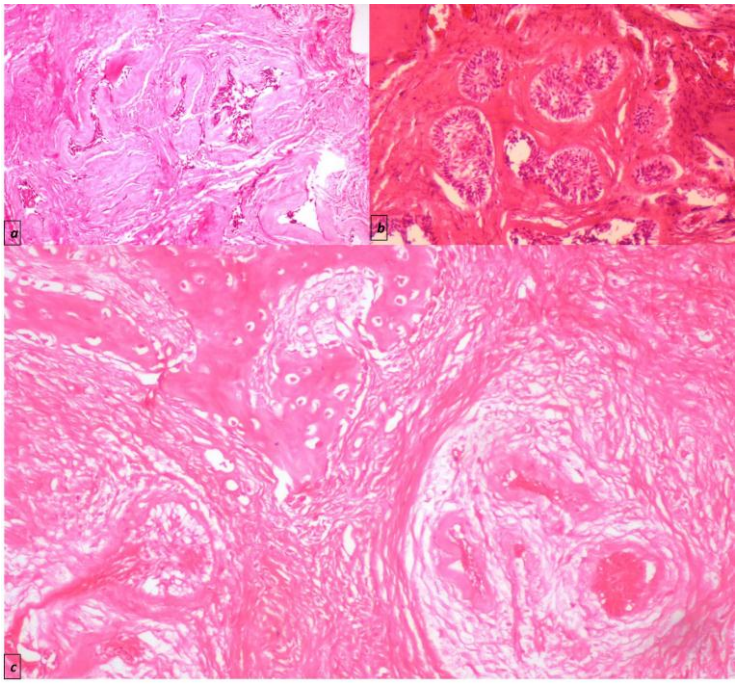

Figure 2: The hematoxylin and eosin stained photomicrograph at x40 showing dense collagen tissue compressing the ameloblastic islands and follicles with squeezing effect (a). At x100 showing typical ameloblastic follicles containing peripheral tall columnar ameloblast-like cells with reverse polarity of the nucleus and central stellate reticulum-like cells surrounded by desmoplastic stroma (b). At x200 the ameloblastic follicles surrounded by desmoplastic tissue and osteoplastic bone showing prominent osteoblastic rimming with plump osteocytes in the matrix (c).

In our present case, histopathologically, marked stromal desmoplasia is one of the most important features, along with the presence of compressed ameloblastoma like follicles lead to a diagnosis of desmoplastic ameloblastoma.[9] Presence of classical follicles with peripheral palisading of ameloblast-like cells with reverse polarity of nucleus, containing central core of stellate reticulum-like cells, and few areas of acanthomatous and basaloid differentiation point towards a “hybrid lesion” of ameloblastoma. Evidence of marked osteoplasia in such a lesion that is seen in our case is extremely rare and not reported in great numbers.

In the present case, a relation is seen between desmoplastic ameloblastoma, “hybrid lesion” and osteoplastic activity. Behavior of odontogenic epithelium with stromal connective tissue can vary from minimal to marked inductive changes as described by Spouge.[8] The inductive activity may include classical desmoplasia and in some instances, osteoplastic activity.[4] The osteoplastic activity in the stroma may be induced by the tumor cells resulting in bone formation in a pattern very similar to the formation of desmoplasia. The stimuli which cause stromal fibroblasts to cause desmoplasia may also lead to differentiation of stromal mesenchymal cells to form osteoblasts causing osteoplasia.[4] The hybrid lesions have a tendency to have abundant stroma and desmoplasia, especially, when occurring in the tooth bearing areas.[7] The present lesion is in a tooth bearing area of the bone, showing a marked tendency of desmoplasia and osteoplasia, though the etiology for the same is not known. Very few such cases are reported leading to

insufficient knowledge about clinical, radiographic, histological features and behavior of the tumor.[10]

Seeing the array of histopathological findings in desmoplastic ameloblastoma, we conclude that there can be three variations.

1. **Classic:** Shows typically compressed follicles with marked stromal desmoplasia.
2. **Osteoplasia:** Tumor shows features of stromal desmoplasia and osteoplasia along with compressed odontogenic follicles.
3. **Hybrid lesion with or without osteoplasia:** Tumor shows classic ameloblastic follicles with follicular, acanthomatous, and basaloid patterns, with presence of marked stromal desmoplasia with or without osteoplasia.

The intention is to elucidate, if a particular histological pattern leads to a different behavioral patterns.

Conclusion

Desmoplastic ameloblastoma is a proven separate entity of ameloblastomas. A hybrid lesion of desmoplastic ameloblastoma with osteoplastic pattern needs further examination of existing cases and inclusion of new cases to understand the behavior and further clinical implications of the same. Radiographically, desmoplastic ameloblastoma seldom suggests the diagnosis of ameloblastoma and usually resembles a fibrous-osseous lesion because of its radiolucent and radioopaque appearance. Hence, biopsy should be taken for confirmation as prognosis, treatment and biologic behaviour vary. Therefore, the clinician must be aware of these lesions and should keep the patient for follow–up to check for recurrence. In our present case there was no recurrence after a regular check-up for 26 months.

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