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Case Report

Histological and clinicoradiographic evaluation of desmoplastic ameloblastoma

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ABSTRACT

A rare histologic variant of ameloblastoma, desmoplastic ameloblastoma is an odontogenic neoplasm that is challenging to diagnose. According to the case report, a 37-year-old female patient with history of intermittent pain and a hard swelling in the maxillary anterior region that measured 10×12 mm is discussed here. This variant frequently occurs in the anterior maxilla, despite the fact that ameloblastomas are typically found in the mandible. Following radiographic and clinical evaluation, surgical enucleation was carried out, and histopathological analysis was completed with the aid of H and E staining. In order to diagnose this uncommon form of ameloblastoma, we are attempting to shed light on the clinico-radiographic and histopathological features of desmoplastic ameloblastoma.

Keywords: Desmoplastic ameloblastoma, Odontogenic tumor, Desmoplasia

INTRODUCTION

Ameloblastoma, the second most common benign odontogenic tumor, is characterized by its locally aggressive, destructive, and invasive behavior.[1-3] Ameloblastoma presents clinically as a solid or multicystic, unicystic, and extraosseous. It usually manifests as a multilocular or unilocular radiolucency on radiography.

Ameloblastoma refers to various histological patterns, with the follicular and plexiform types being the most prevalent, followed by the acanthomatous and granular cell variants.[4] Desmoplastic, basal cell, clear cell, keratoameloblastoma, and papilliferous keratoameloblastoma are uncommon variations.

"Ameloblastoma with pronounced desmoplasia," the first thorough report on the condition in the English literature, was the term Eversole used in 1984 to describe three cases of the desmoplastic variant of ameloblastoma.[5] The rare variation of odontogenic tumors known as desmoplastic ameloblastoma is included in the World Health Organization's (WHO) classification system.[6] Two widely recognized histologic forms of DA are DA with osteoplasia (12.0%) and simple DA (88.0%0).[7]

DA's radiographic features are nonspecific and can be mistaken for either malignant tumors or fibro-osseous lesions.[8] Ameloblastoma of desmoplatic variant shows more aggressive spread as compared to other forms of ameloblastoma. This might be due to its tendency to enlarge and its peculiar anatomical position in the anterior maxilla, which may result in an early invasion of neighboring bone and structures.[9]

In order to better understand this variation in odontogenic tumors, this article will highlight a case of ameloblastoma that has the unique clinical, radiographic, and histological features associated with DA.

CASE REPORT

A 37-year-old woman was referred to the periodontology department after presenting to the oral medicine and radiology department of RUHS College of Dental Sciences in Jaipur, Rajasthan, with the primary complaint of swollen gums. Her left maxillary anterior region had developed an asymptomatic swelling of spontaneous origin during the previous 20 to 25 days. Which was progressively growing until it reached its current size. She reported having intermittent pain but denied any bleeding or change in her senses. There was no significant traumatic, dental and medical history. The intraoral examination revealed an enlarge mass, approximately 10×12 mm in size, extending from the distal surface of left lateral incisor to the mesial surface of left canine buccally. Buccopalatal expansion of the involved site was not evident. The mucosa over the swelling appeared normal (Figure 1).

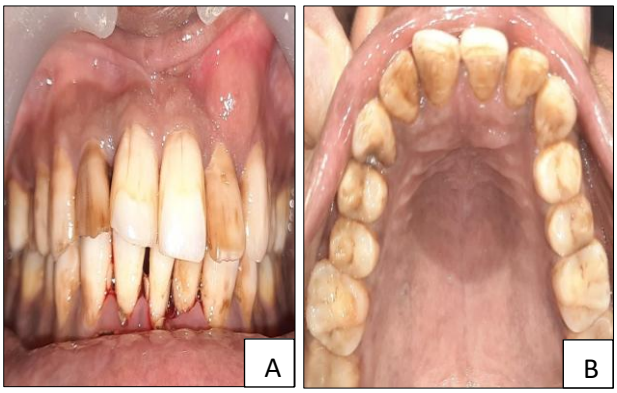

Figure 1 (A and B): Clinical front view of the patient at baseline showing gingival overgrowth between maxillary left lateral incisor and canine and clinical picture of palatal view at baseline.

On palpation, the swelling was bony hard, non-tender and was not easily compressible nor fluctuant. There were no discernible mobility and the teeth in the diagnosed area were not sensitive to percussion. Electric pulp vitality tests showed that all of the teeth next to each other were vital. There were neither lymphadenopathies nor fistulae.

A radiographic analysis with help of cone beam computed tomography (CBCT) scan of the maxilla showed a unicystic/unilobulated, well defined radiolucency with ill-defined radiopaque margin of the lesion that was approximately 17.2 mm (buccopalatal/anterioposteriorly) ×17 mm (apicocoronal/superioinferiorly) in size and extended from the distal surface of the #22 to the mesial surface of the #23. The lamina dura surrounding the affected teeth was lost. The lesion forced the roots of the #22 and #23 move apart, but there were no signs of root resorption (Figure 3). The radiograph revealed that the maxillary sinus floor was intact (Figures 2 and 3). After clinical and radiographic evaluation, globulomaxillary cyst was considered as provisional diagnosis. Lateral to the midline, it manifests intraorally as a soft-tissue enlargement of the maxillary anterior mucolabial fold. On maxillary anterior radiographs, it appears as a "inverted pear-shaped radiolucency" between a maxillary lateral incisor and the neighboring canine. The globulomaxillary cyst often causes the roots of nearby teeth to diverge. As globulomaxillary cyst is a developmental cyst that origin from entrapment of non-odontogenic epithelium of globulomaxillary suture.[10]

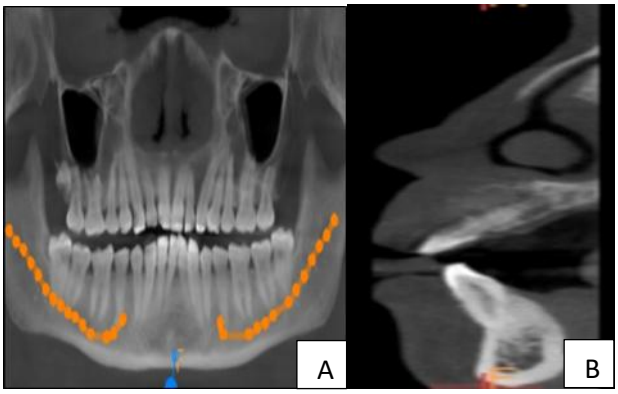

Figure 2 (A and B): Panoramic view of CBCT scan showing interradicular radiolucency and divergence of roots of #22 and #23 and paraxial view showing unilocular radiolucency.

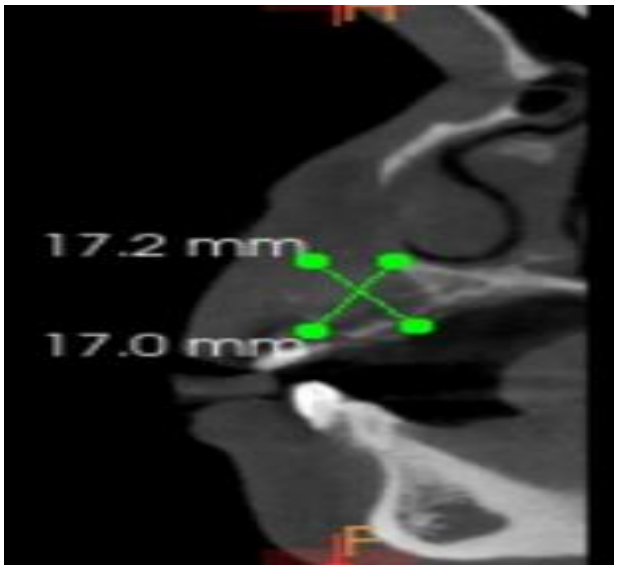

Figure 3: Measurement of the lytic lesion of CBCT Scan.

The lesion could not be aspirated, and a complete hemogram showed values that were normal. Thus, enucleation surgery was planned.

Surgical procedure

Local infiltration of 2% lignocaine hydrochloride with adrenaline 1:80,000 was used to anesthetize the surgical site (from #21 to #24) both labially and palatally. A

crevicular incision and an interdental incision were made using a surgical blade number 15. From the mesial surface of #21 to the mesial surface of #24, vertical releasing incisions were created on both sides of the surgical site, and a complete thickness (Figure 4).

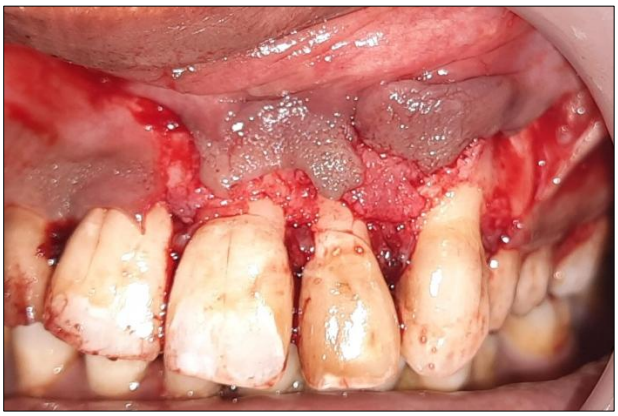

Figure 4: Reflection of full thickness mucoperiosteal flap at surgical site.

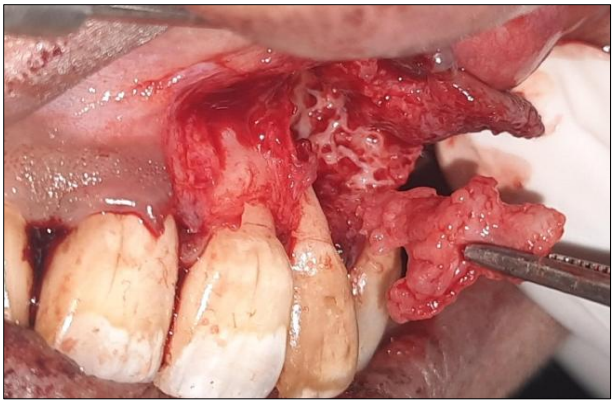

Figure 5: Surgical enucleation of lesion.

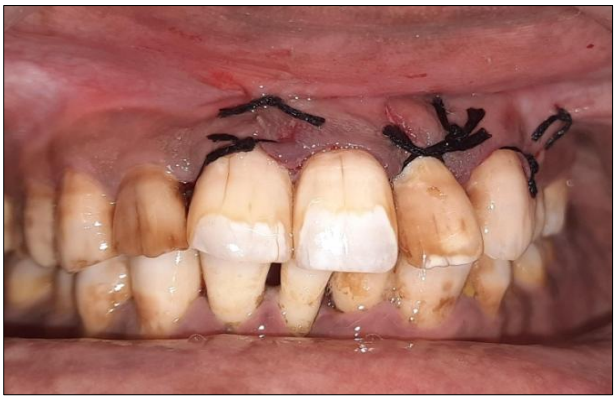

Figure 6: Flap was sutured back to original position.

A thorough enucleation was done and it was also observed that the cortical bone above it was thinning, with few, irregular internal trabculae and no discernible extension into the sinus after that the reflected mucoperiosteal flap was sutured with 3-0 black silk suture back to the original position (Figures 5 and 6). The surgical specimen of the enucleated cystic lesion sent for histopathological analysis. Patient was recalled after 1 week for suture removal and re-evaluation at 1 month post-operatively shows satisfactory healing outcome (Figure 7).

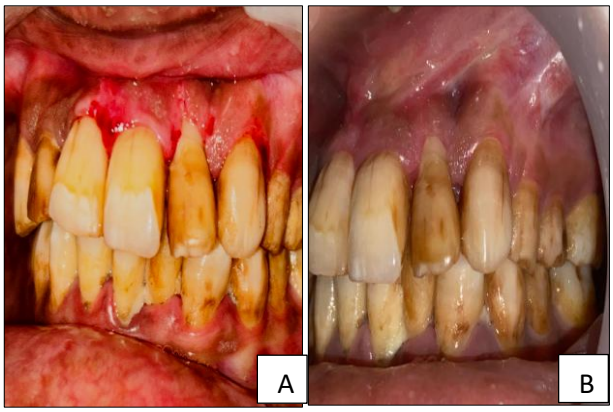

Figure 7 (A and B): Post operative clinical pictures at 1 week and 1 month shows satisfactory clinical outcome.

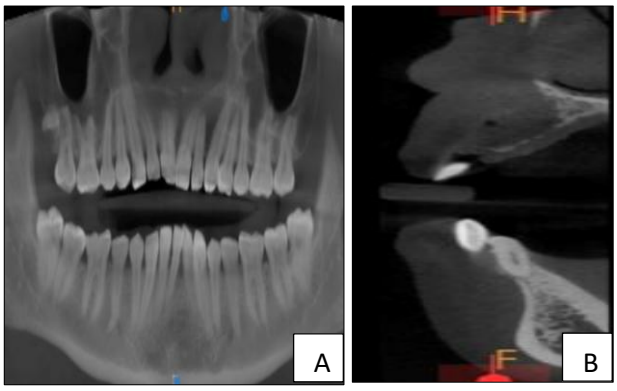

Figure 8 (A and B): Post-op CBCT scan panoramic view and paraxial view reveals that the radio-opaque lining of the cyst has disappeared.

Immediate post-op CBCT scan was performed in order to rule out whether the cystic lining has been properly enucleated (Figure 8).

Histopathological examination

The confirmatory diagnosis was made after histopathological analysis of biopsy specimen which appears as desmoplastic variant of ameloblastoma. When examined under a microscope, the following characteristics are typically seen: the most consistent and distinctive feature of stromal desmoplasia is its moderately cellular, fibrous connective tissue with more collagen; islands of the epithelial component in various shapes; cuboidal cells in the peripheral layer; and spindle-shaped or polygonal epithelial cells made a hypercellular central area (Figures 9 and 10).

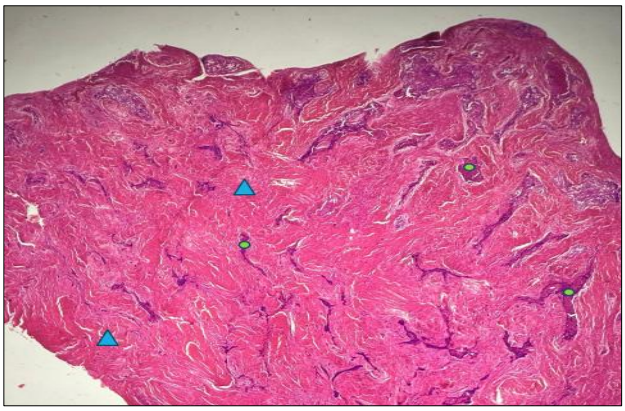

Figure 9: Histopathological pictures, microscopic features after H and E staining shows moderately cellular fibrous connective tissue strauma and odontogenic epithelial islands; at 4X magnification.

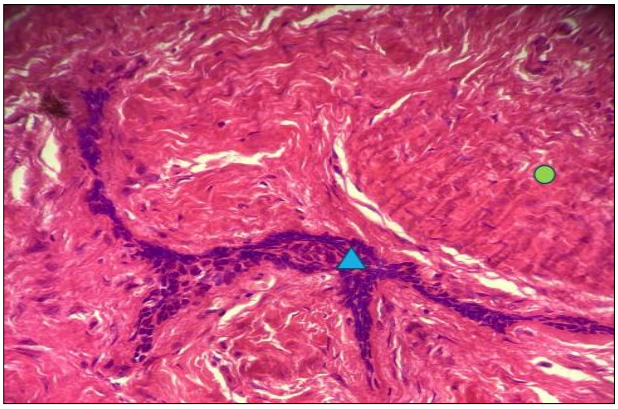

Figure 10: At 40X magnification strands of epithelial island.

These typical characteristics mentioned in the literature aligned with our case. DA can occasionally show plexiform or classic follicular ameloblastoma zones scattered throughout; these have been dubbed "hybrid lesions".[11]

Differential diagnosis

A histological differential diagnosis could include ameloblastic fibroma, odontogenic fibroma, and squamous odontogenic tumor. Finding the typical ameloblastic areas is essential for an accurate diagnosis of the desmoplastic variant of ameloblastoma, which may necessitate additional tissue examination or a repeat biopsy.

DISCUSSION

DA is still one of nature's mysteries because there are so few case reports of it. In majority of the cases, most noticeable clinical manifestation is a painless swelling or a bony expansion. The percentage of DA ranges from 0.9% to 12.1% of all the ameloblastomas. The age and gender predilection are comparable to those of other ameloblastomas, with the mean age at the initial presentation being 42.3 years (range: 17–70 years). The relative frequency of DA appears to be slightly higher in Asian populations, according to data from various geographic regions, suggesting a biogeographical pattern.

It is hypothesized that the periodontal membrane of the associated tooth is where DA originates. Furthermore, it has been suggested that the epithelial rests of Malassez within the periodontal ligament may serve as the origin of DA. Here it was noted that the lamina dura and periodontal ligament space of adjacent lateral incisor was disintegrated.

Most desmoplastic lesions often develop in the anterior or premolar regions of the jaws, with approximately half occurring in the maxilla. The unicystic or classic forms of ameloblastoma, on the other hand, are typically found in the mandibular posterior region. Because the maxillary sinus and important structures are so close together, maxillary lesions are more concealed than mandibular tumors. Furthermore, the extremely thin cortical bone of maxilla acts as a fragile barrier to stop tumors from spreading. As a result, maxillary ameloblastomas tend to spread earlier and more rapidly than their mandibular counterparts.

DA typically shows up on radiograph as a mixed radiolucent and radiopaque lesion, which can occasionally resemble a benign fibro-osseous lesion. Osseous metaplasia within the dense fibrous septa defining the lesion, is the cause of the mixed radiographic appearance, not a mineralized product produced by the tumor.

According to Philipsen et al, radiographically ill-defined borders suggest an infiltrative process with a high likelihood of recurrence. The radiographic features of DA are nonspecific and may mimic those of fibro-osseous lesions, resembling conditions such as fibrous dysplasia or chronic sclerosing osteomyelitis, and, when well-circumscribed, may appear similar to an ossifying fibroma.[12] Additionally, the absence of the usual ameloblastoma findings made the current case difficult to diagnose. The appearance of the radiograph led us to suspect a fibro-osseous lesion.

Because not all of the epithelial clusters exhibit the characteristic palisading layer of ameloblastoma, the tumor may histologically be confused with another odontogenic tumor, particularly if the biopsy sample is small. Odontogenic fibroma may be mimicked by regions of desmoplastic stroma that contain only thin strands of epithelial cells. The distinctions between these two tumor's clinical characteristics and approaches to treatment are what make them significant.[13]

Because the desmoplastic form of ameloblastoma may contain a fibrotic stroma, the squamous metaplasia observed in some locations may resemble the palisading layer of the tall columnar cells if it cannot be differentiated.

Ng and Siar et al used a variety of immunohistochemical techniques to show that the tumor cells of DA expressed desmin and S-100 protein in different ways. In a similar vein, connective tissue stroma from DA has been shown to strongly react favourably to collagen type VI. Scar tissue was ruled out, as the findings suggested active de novo synthesis of extracellular matrix proteins.[14]

The best course of treatment for DA is still unclear due to a lack of knowledge about its biologic behavior and prognosis. Resection remains the most common treatment for DA, although curettage or enucleation has been employed in selected cases. In this case, surgical enucleation was the recommended treatment. Curettage, on the other hand, results in tumor islands within the bone that subsequently recur. According to Keszler et al, the recurrence rate of the desmoplastic form of ameloblastoma was higher (21.4%) than that of the other forms (10.1%).[15]

CONCLUSION

DA is distinguished by particular histological, imaging, and clinical characteristics. Long-term monitoring and more thorough analysis are necessary for a proper understanding of such cases. Any lesion, from a simple abscess to any fibro-osseous lesions or neoplastic growth presenting in the anterior maxilla or mandible, should have DA included as a differential diagnosis by the clinician due to the neoplasm's unusual presentation. A definitive diagnosis requires histopathological analysis.

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