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Hemangiomatous Ameloblastoma: Surgeon's Perspective and A Report of Two Cases

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Abstract

Ameloblastoma is a benign tumor of jaws with locally destructive nature and hemangiomatous ameloblastoma (vascular ameloblastoma) is its rare variant. Owing to a very few reported cases and a lack of follow-up, the behavior and prognosis of hemangiomatous ameloblastoma is uncertain.This article presents two case reports of hemangiomatous ameloblastoma with an emphasis on its diagnostic features and surgical considerations.

Keywords: Hemangiomatous, Diagnostic, Plexiform, Acanthomatous, Desmoplastic

Introduction

Ameloblastoma is a slow-growing solid or cystic benign odontogenic neoplasm of epithelial origin. It is the second most common odontogenic tumours (11-13%). The ameloblastoma is an entity of interest owing to its polymorphic nature, with various clinical and histopathological variants. According to WHO (2017), its clinical variants include conventional, unicystic, peripheral (extra-osseous) or malignant type. Histopathological variants include follicular, plexiform, acanthomatous, desmoplastic, granular cell and basal cell

types[1]. The hemangiomatous ameloblastoma was described, in the literature, as an ameloblastoma in which part of the tumour showed presence of blood-filled spaces or space containing large endothelial-lined capillaries. It was originally introduced by Kuhn by his first report of extremely vascular form of ameloblastoma in 1932, a combination of hemangioma and adamantinoma as a central jaw tumour [2].Lesions with similar presentation in literature are termed as angiomatous ameloblastoma, hemangio-ameloblastoma, adamantino-hemangioma, vascular ameloblastoma, ameloblastic hemangioma and collision tumour. On the other hand, Smith in 1966did not consider it a distinct histologic entity due to variable blood supply to this tumor [3]. Diagnosis and surgical treatment of two cases of hemangiomatous ameloblastoma are reported in this article.

CASE 1

A 42-years old female patient visited the Department of Oral and Maxillofacial Surgery of our institution with a chief complaint of gradually enlarging painless swelling on left side of facefor 6 months. The medical history was non-contributory. On examination, extraorally, there was a single, smooth, firm, non-fluctuant swelling on left side of lower face. Intraorally, a swelling in left sided mandibular region, obliterating the buccal vestibule was present. Paresthesia was absent.

Cone Beam Computed Tomography was done and it revealed a single septate multilocular radiolucency involving left side of mandible, extending from premolar region upto sigmoid region and inferiorly extended till lower border of the jaw. The axial section revealed extensive buccolingual expansion of the jaw on left side. Incisional biopsy was done uneventfully under local anaesthesia and the specimen was sent for histopathological examination. The microscopic histopathological examination revealed features of hemangiomatous ameloblastoma [Figure 1].

The segmental resection of mandible on left side with disarticulation, taking a safe margin into consideration, was performed under general anaesthesia [Figure 2]. During surgery, due to high vascularity of the lesion, local hemostatic measures were used including agents like absorbent gelfoam. Hypotensive anaesthesia was maintained during the complete procedure. The reconstruction was planned but could not be done due to patient's refusal. The postoperative healing was uneventful. No recurrence was reported after a follow up period of two years.

CASE 2

A female patient, aged 45 years, reported to the Department of Oral and Maxillofacial Surgery of our institution with a complaint of a small swelling in right side of mouth for 8 months. The swelling was sudden in onset and gradually increased in size. There was a history of extraction of the regional teeth but rest of medical history was insignificant. The swelling was discharging in nature and was associated with intermittent mild pain. On clinical examination, there was a firm, non-tender swelling on right side of face in the area of lower jaw. Multiple missing teeth were present intraorally. Discharge on palpation was present.

Panoramic radiograph showed a well demarcated, well corticated multilocular radiolucent-radiopaque lesion extending from incisor region of left side to molar region of right side, not involving the inferior border of mandible. Magnetic Resonance Imaging revealed slow flow, serpentine like vascular structures in right side of mandible in the area of lesion. The lesion showed expansion more on buccal side than lingual side [Figure 3].

Incisional biopsy was performed under local anaesthesia and was uneventful. The histopathological examination of the specimen was done and the presence of vascular spaces was evident [Figure 4]. It was confirmed to be a vascular variant of ameloblastoma. The surgical plan for this lesion was surgical excision with peripheral ostectomy under general anaesthesia but patient was reluctant for the same. So, enucleation of the lesion with curettage was done under general anaesthesia [Figure 5]. The lesion was vascular but the surgery was uneventful without significant bleeding. The postoperative healing was uneventful and the bony healing was evident radiographically during follow-up period. No recurrence was reported after a follow-up of twenty months.

Legends Figures

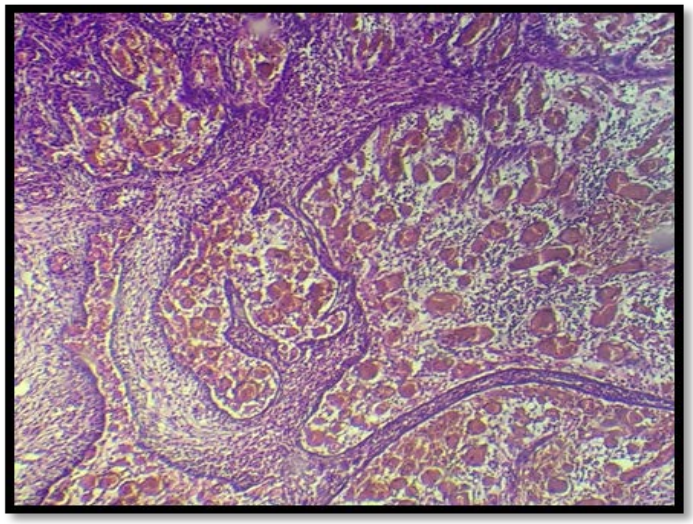

Figure 1: Photomicrograph showing vascular spaces

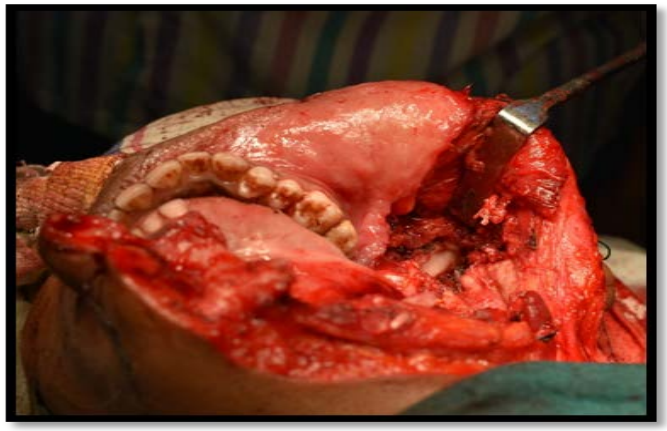

Figure 2 : Segmental resection of mandible with disarticulation performed

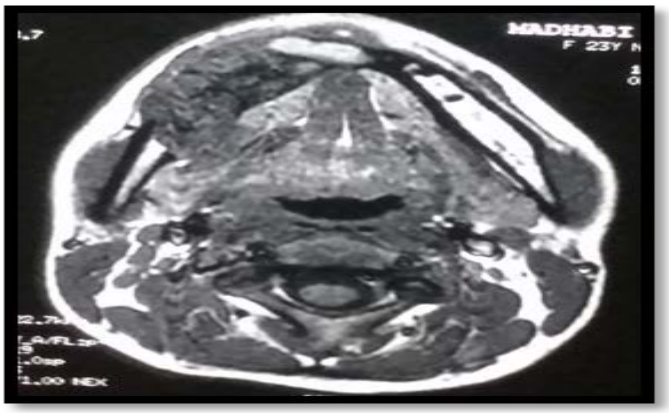

Figure 3: Axial MR image depicting the nature of lesion on right side of mandible

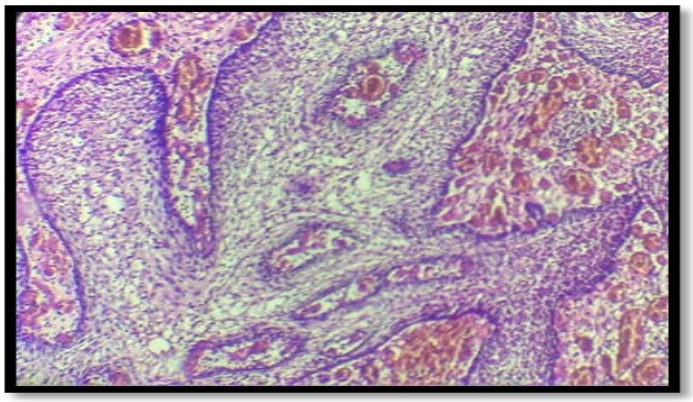

Figure 4: Photomigrograph showing ameloblastomatous epithelium with numerous endothelium lined vascular spaces

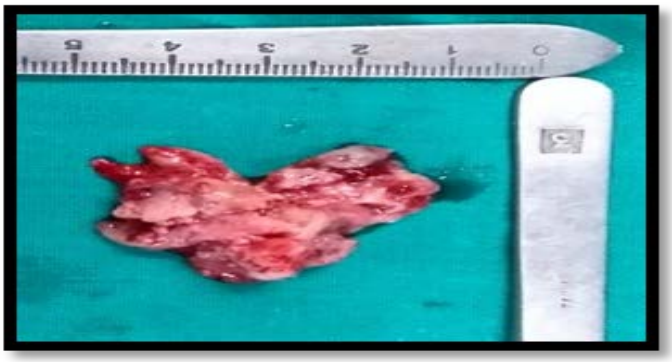

Figure 5: Specimen after enucleation of the lesion

Discussion

Hemangiomatous ameloblastoma (HA) has experienced a notorious timeline of events related to its existence as a

separate entity. After first reported case of HA by Kuhn (1932), Thoma, in 1944, listed it in classification of benign odontogenic tumors of jaw[4]. In 1957, Lucas rejected its designation of hemangioameloblastoma owing to no vasoformative activity[5]. In 1961, it was declared as a collision type of tumour[6]. Its existence was again rejected in 1968. Its struggle for a specific standardised terminology is still going on. Ameloblastoma accounts for about 1% of all oral tumors. Its vascular variant is very rare with less than twenty cases reported till date. HA is more prevalent in middle age persons with mean age of occurrence being 30 years. Hemangiomatous ameloblastoma presents with no sex predilection, occurs commonly in posterior region of mandible, with very rare occurrence in maxilla. In the present case reports, one patient was 42-years old and the age of the other patient was 45 years. Both the patients were females and the lesion were present in posterior mandible in both the cases.

The pathogenesis of vascular component is still unclear with various proposed theories like: angiogenesis during development of tumor; representation as a collision tumor or; as a separate neoplasm; hamartomatous malformation; as a result of traumatic incident; as a secondary change[7]. Among the reported cases in this article, there was a history of dental extraction in one case.

The peculiar radiographic features of HA, as observed on Computed Tomograms, include its soft tissue contents, expansile nature and proliferative endosteal reaction, which necessitates its differentiation from fibro-osseous lesions. Fibro-osseous lesions like ossifying fibroma show similar features but, in contrast to HA, have a surrounding radiolucent line corresponding to fibrous capsule. Magnetic Resonance Imaging of Hemangiomatous Ameloblastoma presents with a vascular stroma containing large interstitial spaces; serpentine-like vascular structures, demonstrating the angiomatous nature of HA[8]. In this case, MRI presented with similar features. Histopathologically, it consists of an ameloblastoma with a prominent vascular component, which is missing in conventional variants[9]; same as seen in both the present cases.

Segmental resection was done in 42.8% of all reported cases; while enucleation was done in other cases.The use of carnoy's solution following enucleation is also reported in one case [10]. There was uneventful postoperative healing reported in all the cases; with no episode of recurrence.Significant bleeding episodes are reported in 14.3% of cases. In our both cases, the operating surgeons didn't experience any inadvertent bleeding episode, however, used local hemostatic agents and hypotensive anaesthesia as a precaution.

Conclusion

Hemangiomatous ameloblastoma is a rare entity and should be considered as a differential diagnosis for bony lesions of maxillofacial region.

Since there are a very few reported cases of Hemangiomatous Ameloblastoma, there are still no specific surgical guidelines established for HA. A specific treatment protocol for HA may help to avoid any fatal hemorrhagic episode at the time of a surgery.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms.

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