Hemangioameloblastoma: A Rare Presentation

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ABSTRACT
The hemangioameloblastoma is described as an ameloblastoma in which part of tumor contains spaces filled with blood or large endothelial-lined capillaries. Variations in histomorphologic pattern do not appear to have a significant bearing on the biologic or prognosis of these tumors. The present case report describes a rare case report of a 32-year-old male patient with intrabony pathology in body of mandible crossing midline, where the clinical and radiographic picture was atypical of ameloblastoma, however on complete imaging and histopathological evaluation, it was diagnosed as hemangioameloblastoma.

Keywords: Hemangioameloblastoma, Endothelial-lined capillaries, Body of mandible.

INTRODUCTION
Ameloblastoma is a benign epithelial odontogenic tumor that usually exhibits aggressive behavior. It expands severely to the cortical bones and may have a high recurrence rate. It also may cause mobility and displacement of the teeth, as well as root resorption.1

Their polymorphic nature is reflected by the variety of recognized histologic patterns with which they may appear. The follicular and plexiform patterns are the main histologic types. Commonly encountered histologic variants are acanthomatous and granular cell types.2 Other less commonly encountered histologic patterns include desmoplastic ameloblastoma,3 basal cell ameloblastoma,4 clear cell ameloblastoma5 and unicystic ameloblastoma.6 The theory of an odontogenic origin for the ameloblastoma is supported clinically by the tumor’s common occurrence in the tooth bearing area and is further reinforced by the finding of Spouge that one in every three such tumors are mural proliferations in intimate association with the reduced enamel epithelium of dentigerous cysts.7 Variations in histomorphologic patterns do not appear to have a significant bearing on the biologic behavior or prognosis of these tumors, with the possible exceptions of unicystic and desmoplastic types.3,5

The hemangiomatous ameloblastoma (HA) was originally described as an ameloblastoma in which part of the tumor contained spaces filled with blood or large endothelial-lined capillaries.7 Lesions with similar histologic features that probably represented the same entity were documented in the early literature as adamantinohemangiomas,8,9 ameloblastic hemangiomas10 and hemangioameloblastomas.11 In 1966, Smith12 suggested that the HA should not be regarded as a separate histologic pattern because the blood supply to these tumors was variable and other circumstances may have affected the number and size of the blood vessels associated with them.

The origin of the vascular component of the HA is not completely resolved, and the histologic and radiologic features differ from those of the accepted types of ameloblastomas. Additional published cases will provide data for a complete clinical and prognostic profile of this lesion. This article presents an ameloblastoma with clinical, radiologic and histologic features consistent with those of an HA.

CASE REPORT
A 32-year-old male patient was referred to our department with gradually enlarging asymptomatic swelling in the posterior region of left mandible since 6 months and he had been taking anti-inflammatory drugs for swelling. He had given history of similar swelling previously 10 years back for which he was operated along with extraction of left first premolar. Patient did not present any other sign and symptoms. There was no relevant medical and family history. Patient was not having any tobacco habits; rather he was having normal lifestyle.

During general examination, his gait, build and posture were normal no any physical deformity was found. Vital signs were in normal range. There was no cyanosis, clubbing or icterus seen. In extraoral examination face appeared to be bilaterally symmetrical as it was not showing any evident swelling. Vital signs were in normal range. There was no cyanosis, clubbing or icterus seen. In extraoral examination face appeared to be bilaterally symmetrical as it was not showing any evident swelling. However, on palpation there was a firm, tender swelling was present on left side of body of mandible. Overlying skin appeared normal with no increase in temperature but there was a scar of previous surgical procedure near inferior border of left side of mandible. Lymph nodes were nonpalpable. TMJ movements were normal. No other signs were noted.
In intraoral examination, a well-defined, localized, lobulated swelling was seen in the left side of mandible extending from left canine to left first molar in buccal vestibule. Overlying mucosa was reddish. There was no any discharge through swelling. Mandibular first premolar was missing which was extracted 10 years earlier. Mandibular second premolar was having grade III mobility (Fig. 1). On palpation, the swelling was tender and expansion of both buccal and lingual cortical plates was noted. There was no change in temperature of involved mucosa. Vitality tests on teeth in third quadrant are however normal.

Based on these clinical findings, we arrived on provisional diagnosis as ameloblastoma, central giant cell granuloma, ossifying fibroma, odontogenic keratocyst, residual cyst.

Intraoral periapical radiographs showed a large radiolucent lesion involving left side of mandible, extending anteriorly mandibular anterior teeth region up to posteriorly mesial aspect of second molar. Lesion appeared to be crossing midline. In anterior teeth region, lesion appeared to be mixed radiolucent and radiopaque areas giving. While in posterior teeth region, a well-defined radiolucency with no corticated border was seen. Resorptions of root of premolar and distal root molar were seen.

Occlusal, panoramic view (Fig. 2) showed a well-defined, multiloculated radiolucency involving left hemimandible, crossing midline extending posteriorly up to mesial aspect of second molar. Integrity of inferior border appeared to be maintained and no pathological fractures were seen.

Computer tomography (CT) (5 mm axial and coronal slices, bone and soft tissue windows) (Fig. 3) oriented on lower border of mandible displayed well-defined abnormal hypodense multilocular lesion involving the left hemimandible measuring approximately 2.7 × 3.2 × 1.6 cm crossing midline and extending posteriorly up to molars. The integrity of alveolar canal was maintained. There was no abnormal associated soft tissue component or pathological fractures seen.

Radiographic differential diagnoses were made as an ameloblastoma and central giant cell granuloma. After biopsy, (Fig. 4) microscopic examination revealed odontogenic epithelial lining which was proliferating in the lumen in the form of plexiform pattern. Odontogenic islands were seen in the connective tissue with homogenous eosinophilic calcifications. The stroma showed numerous engorged blood vessels with blood elements and also cavernous spaces. Features were suggestive of hemangioameloblastoma. So the final
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Diagnosis of hemangioameloblastoma of mandible was made and surgical resection of this tumor was done and its follow-up was also done.

DISCUSSION

Various theories have been put forth to explain the pathogenesis of the vascular component in ameloblastomas. During amelogenesis, many capillaries are associated with the outer enamel epithelium. They furnish the profuse blood supply necessary for enamel completion. It is probable that in the HA these blood vessels are abnormally induced to become part of the tumor. It is also said that excessive stimulation of angiogenesis during tumor development, by inductive influences such those that occur during odontogenesis or by other factors, may result in the overgrowth of the vascular elements in the odontogenic ectomesenchyme or in the adjacent connective tissue.

Alternatively, a traumatic incident such as a tooth extraction may provide a stimulus required for proliferation of epithelial cell rests in the periodontal ligament and subsequent tumor development. Tissue damage is usually followed by repair and this involves the formation of the granulation tissue in which proliferating endothelial cells and new capillaries are prominent. A disturbance in the repair of neoplastic odontogenic tissue may result in excessive granulation tissue formation or the development of an abnormal vascular component.

It has been suggested that the HA represents a collision tumor. In this type, two separate tumors grow in the same area and collide, and the tumor elements intermingle.

Smith regarded the HA as histologically similar to one of the other recognized types of ameloblastoma and not as a distinct histologic entity. He thought the blood supply to these tumors was variable and that circumstances other than the number and size of the vessels influenced the blood supply. Whether the vascular component of the HA is part of the neoplastic process, represents a separate neoplasm, or is a hamartomatous malformation has not been satisfactorily resolved.

The HA observed here differs histologically and radiologically from a conventional ameloblastoma. Histologically, it consists of an ameloblastoma with a prominent vascular component while its conventional radiologic features are nonspecific.

CT scan clearly demonstrates cystic features in this tumor, such as its expansile nature and soft tissue contents.

It is generally accepted, that the mural or unicystic ameloblastoma represents a distinct entity that occurs during the first two decades of life. The diagnosis is made histologically, and growth follows a more benign course. At radiography these lesions appear as a well-defined, well-corticated radiolucency resembling a dental cyst.

The biologic behavior is thought to be similar to that of the conventional ameloblastoma, but because few cases have been reported, the pathogenesis and clinical features are not yet fully understood and biologic behavior cannot be predicted.

CONCLUSION

There have been very few cases reported in literature of HA. The case reported here is of a 32-year-old male patient, with left mandibular premolar-molar region showing buccal and lingual cortical plate expansion. Radiographically, it showed unilocular well-defined radiolucency with root resorption of associated teeth. Based on histopathological findings the lesion was diagnosed as HA. This case clearly demonstrates the distinctive histologic pattern and special imaging features of HA.

REFERENCES