CASE REPORT

Hemangiomatous Ameloblastoma: A Rare Variant

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ABSTRACT

Ameloblastoma is a true neoplasm of enamel organ type tissue. It is the most common odontogenic neoplasm with more frequency in the mandible. A 20 years old male patient presented with a swelling in the right side of the mandible of 10 months duration. Orthopantomograph revealed multilocular radiolucency extending from the region of 46 to the condyle. Incision biopsy revealed features of plexiform ameloblastoma. Numerous vascular spaces of varying size were seen throughout the stroma. Excision biopsy also revealed similar findings. Based on these findings, a diagnosis of hemangiomatous plexiform ameloblastoma was made. Hemangiomatous ameloblastoma (HA) is still a controversial entity, with some pathologists ruling it out as a separate lesion. This paper discusses the possibility that HA might be an aggressive variant of ameloblastoma and reviews relevant literature.

Keywords: Hemangiomatous ameloblastoma, Unicystic ameloblastoma, Ameloblastoma.

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INTRODUCTION

Ameloblastoma is a true neoplasm of the enamel organ type tissue, being a benign odontogenic tumor that develops from epithelial rests of Malassez.[1] It is usually unicentric, nonfunctional, intermittent in growth, anatomically benign, clinically persistent and exhibits aggressive behavior. It represents 1% of all oral tumors and 11% of odontogenic tumors. Its peak incidence is in the 3rd to 4th decades of life and is often associated with an unerupted third molar which can be detected during the course of routine radiography.[2]

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The vast majority of ameloblastomas arise in the mandible, and the majority of these are found in the angle and ramus region and seldom in the maxilla. Furthermore, based on clinical and radiographic characteristics, histopathology and behavioral and prognostic aspects, four subtypes or variants of ameloblastoma can presently be distinguished:[3,4]

i Solid multicystic ameloblastoma (SMA)
ii Unicystic ameloblastoma (UA)
iii Peripheral ameloblastoma (PA)
iv Desmoplastic ameloblastoma (DA), including hybrid lesions.

Variations in the histomorphologic patterns of ameloblastoma do not appear to have a significant bearing on their biologic behavior or prognosis, with the possible exceptions of unicystic, desmoplastic and hemangiomatous ameloblastomas (HA).[5] The HA is a solid multicystic ameloblastoma in which part of the tumor contains spaces filled with blood or large endothelial lined capillaries.[4,6,7]

CASE HISTORY

A 20-year-old male patient was referred to the department of oral and maxillofacial pathology, for the evaluation of a gradually enlarging swelling in the right side of his face, present for almost a year. Patient had visited a private dentist 10 months back for the complaint, and the mandibular right third molar was extracted. However, the swelling did not regress following the extraction.

On examination, a single diffuse swelling of size 4 × 5 cm was noticed in the right lower half of the face (Fig. 1). The swelling was firm, nonfluctuant and painless. Intraoral

Fig. 1: Single diffuse swelling in the right lower half of the face

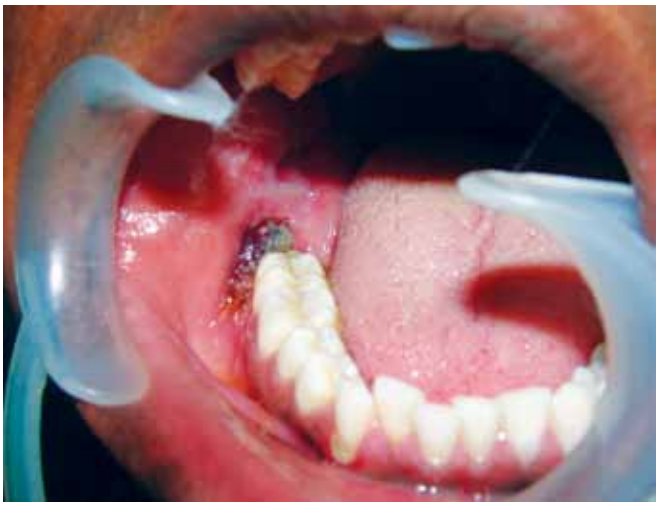

Fig. 2: Intraoral picture shows obliteration of buccal vestibule

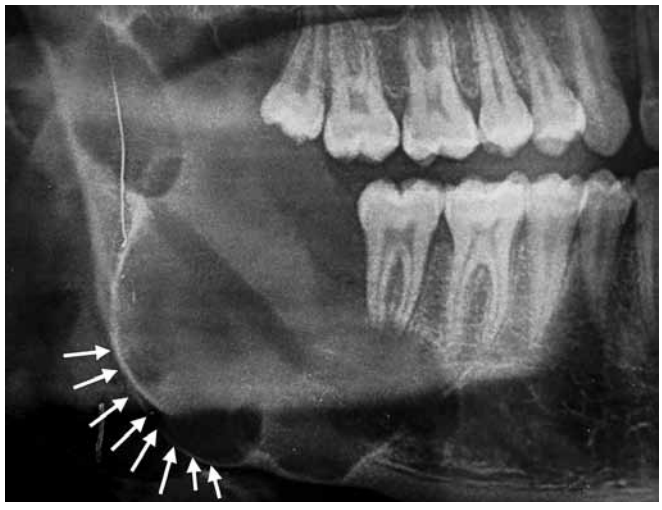

Fig. 3: Orthopantomograph reveals multilocular appearance with 'thinned out' mandible

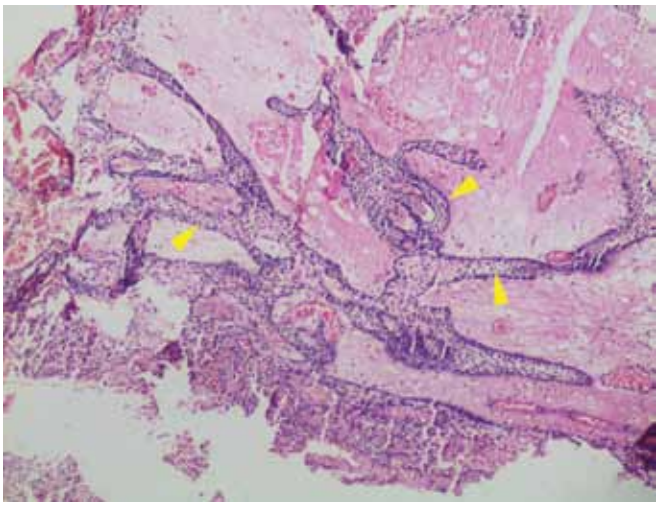

Fig. 4: Photomicrograph showing numerous anastomosing strands of odontogenic epithelium (arrows head) (H&E stain, ×100)

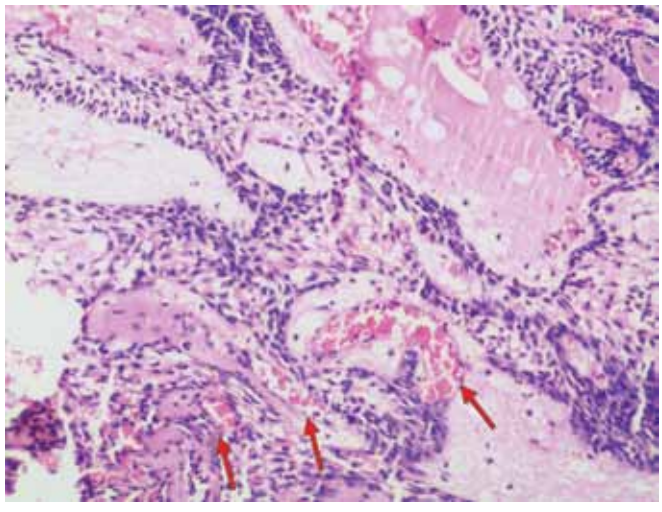

Fig. 5: Photomicrograph showing numerous vascular spaces (arrows) (H&E stain, ×200)

examination revealed a nontender swelling in the right lower quadrant, extending from distal aspect of 46 up to and beyond the retromolar region, with expansion of the cortical plate and obliteration of the buccal vestibule in the region. The swelling involved the entire depth of the lingual vestibule also, thereby obliterating it (Fig. 2). The right submandibular lymph node was palpable, nontender and firm.

IMAGING EXAMINATION

Orthopantamograph revealed a well-defined multilocular radiolucency extending from distal aspect of 46 posteriorly up to the condyle. The lesion appeared predominantly radiolucent with intermingling radiopaque septa giving a soap bubble appearance. The margins of the lesion were scalloped with cortication of antero-inferior and posterior borders. The inferior border of the mandible appeared thinned out in some areas (Fig. 3).

HISTOPATHOLOGICAL EXAMINATION

Sections from the incision biopsy specimen revealed numerous anastamosing strands of epithelium, made up of peripheral cuboidal cells and central cells resembling stellate reticulum. The connective tissue stroma showed bundles of collagen fibres and chronic inflammatory cells. Few blood vessels were also noticed (Figs 4 and 5). A diagnosis of ameloblastoma was made based on these features, and surgical excision of the lesion was advised.

Under general anesthesia, hemimandibulectomy was done (Fig. 6). Sections from the excisional biopsy specimen revealed similar histopathological features as the incision biopsy. In addition, numerous vascular spaces of varying size were also noticed throughout the connective tissue stroma. At places, the vessels were large enough to compress the epithelial strands to almost a single layer of cells. Most vascular spaces were engorged with RBCs, and areas

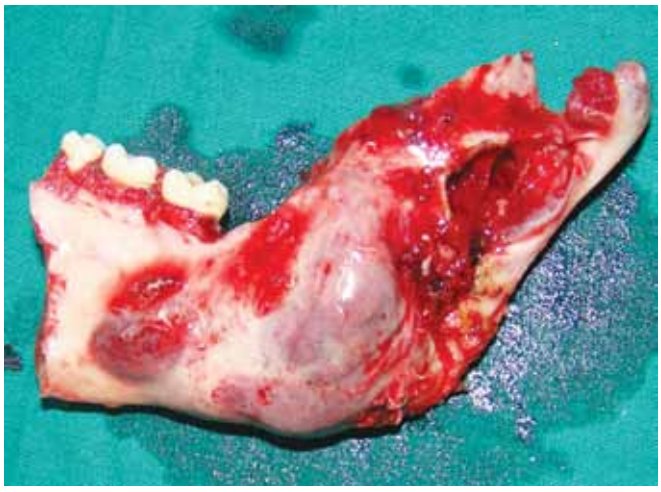

Fig. 6: Excised hemimandibulectomy specimen

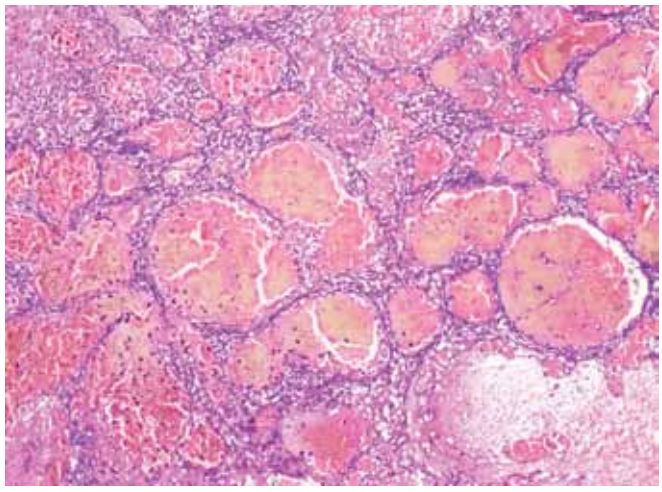

Fig. 7: Photomicrograph showing areas of hemorrhage (H&E stain, ×100)

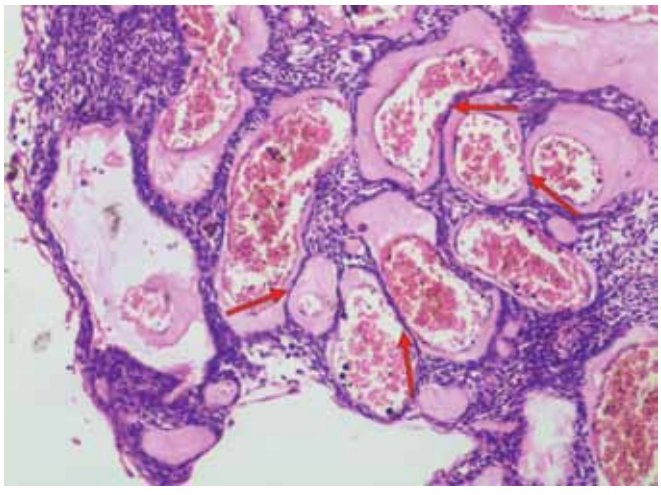

Fig. 8: Photomicrograph showing vascular spaces engorged with RBCs (arrows) (H&E stain, ×200)

of hemorrhage were also seen. Based on these histopathological findings, a final diagnosis of hemangiomatous variant of ameloblastoma was made (Figs 7 and 8).

DISCUSSION

Hemangiomatous ameloblastomas have been reported in lesser frequency than the other classic variants of ameloblastoma, probably because many pathologists do not consider it as a separate variant. Smith considered the HA as being histologically similar to one of the other recognized types of ameloblastomas and not as a distinct histologic entity.[8] The origin of the vascular component of these tumors has always been a source of considerable debate. Various theories have been put forth to explain the pathogenesis of the vascular component in HA.

During amelogenesis, many capillaries are associated with the outer enamel epithelium, providing much needed nutrition to the developing enamel organ. It is probable that in the HA these blood vessels are abnormally induced to become part of the tumor.[9] Excessive stimulation of angiogenesis during tumor development, by inductive influences such those that occur during odontogenesis or by other factors, may also result in the overgrowth of the vascular elements in the odontogenic ectomesenchyme or in the adjacent connective tissue.[4] Trauma to the region resulting in tissue damage, such as due to extraction of a tooth, could also be a stimulus that results in proliferation of epithelial cell rests in the periodontal ligament and subsequent tumor development. Such tissue damage is usually followed by repair and this involves the formation of the granulation tissue in which neoangiogenesis in the form of proliferating endothelial cells and new capillaries is prominent. A disturbance in the neoplastic odontogenic tissue may result in excessive granulation tissue formation or the development of an abnormal vascular component.[9] It has also been suggested that HA represents a collision tumor. In this type, two separate tumors grow in the same area and collide, and the tumor elements intermingle.[5] However, whether the vascular component of HA is part of the neoplastic process, or whether it represents a separate neoplasm, or is a hamartomatous malformation, has not been satisfactorily resolved.[10] In our case, it could be possible that the removal of impacted 48 could have triggered the proliferation of blood vessels in the lesion.

The biologic behavior of HA is thought to be similar to that of the conventional ameloblastoma. A second school of thought considers HA being one of the more aggressive variants of ameloblastoma. However, because only a few cases of HA have been reported till date, the pathogenesis and clinical features are not yet fully understood and biologic behavior cannot be predicted.[10] Surgical complications that can arise as a result of the rich vascularity should be given due importance during treatment planning. It would, therefore, seem prudent to approach a case of HA with caution, especially during the surgical treatment.

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