

Mandibular Haemangiomatous Ameloblastoma with Soft Tissue Infiltration: A Unique Case Report

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ABSTRACT

Hemangiomatous ameloblastoma is a rare histopathological variant of ameloblastoma characterized by prominent vascular proliferation within the tumor stroma. It is often mistaken for primary vascular lesions, resulting in diagnostic difficulty and potential therapeutic challenges. We report a rare case of hemangiomatous ameloblastoma involving the mandible in a patient who presented with a progressively enlarging swelling. Radiographic examination revealed a well-defined expansile radiolucent lesion suggestive of an aggressive odontogenic tumor. Surgical excision was performed, and histopathological evaluation demonstrated typical ameloblastomatous epithelial islands interspersed with numerous dilated blood-filled spaces and proliferating vascular channels, confirming the diagnosis. Notably, infiltration into the adjacent soft tissues was identified, indicating its locally aggressive behaviour. Due to its rarity, limited literature exists regarding the biological behaviour and recurrence potential, mortality rate of this variant. This case underscores the critical role of histopathological examination in differentiating hemangiomatous ameloblastoma from other vascular and odontogenic lesions. Awareness of this entity is essential for accurate diagnosis, appropriate surgical management, and long-term follow-up to ensure public health and to reduce the risk of recurrence.

Keywords: Hemangiomatous ameloblastoma, Mandible, Odontogenic tumor, disease, mortality, Vascular proliferation, Soft tissue infiltration

INTRODUCTION

Ameloblastoma is a benign, locally aggressive tumor that arises from the odontogenic epithelium and is commonly classified into different presentations, such as conventional, unicystic, peripheral, and desmoplastic types.¹ These tumors are most frequently found in the posterior mandible, often presenting as painless, slow-growing masses that are typically diagnosed in middle-aged individuals.² The conventional form is the most common, with six histological variants but rarer variants such as the hemangiomatous ameloblastoma (HA) have also been documented, with distinct pathological and clinical characteristics.³

The term hemangiomatous ameloblastoma was first coined by Kuhn in the year 1932, as a combination of the two terms hemangioma and adamantinoma.⁴ The first ever case was reported by Aisenberg in 1950.⁵ Hemangiomatous ameloblastoma is an uncommon histopathological variant characterized by the presence of well-formed vascular channels within the ameloblastoma tissue, which resembles hemangiomas.⁶ This unusual combination of ameloblastic tissue and vascular structures often leads to diagnostic challenges, as the clinical presentation can mimic other vascular lesions, such as hemangiomas or fibromas.⁷ The vascular component of hemangiomatous ameloblastomas often results in altered clinical features, including sudden swelling, bleeding, or more rapid growth compared to typical ameloblastomas.⁸

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The rarity of hemangiomatous ameloblastomas necessitates a detailed understanding of their clinical, radiographic, and histopathological features for accurate diagnosis. Radiographs and advanced imaging techniques, such as CT scans or MRIs, are essential in identifying the lesion's extent and distinguishing HA from other odontogenic or vascular lesions.⁹ Histopathologically, the defining characteristic of HA is the presence of large, vascular spaces interspersed among the ameloblastic islands, which requires careful examination to avoid misdiagnosis.¹⁰ These vascular features can lead to confusion with other conditions, making a biopsy and thorough histopathological analysis crucial for a definitive diag-

nosis.¹¹

Treatment of hemangiomatous ameloblastomas typically involves surgical resection, similar to other variants of ameloblastoma. However, the presence of the vascular component may necessitate special consideration during surgery to prevent excessive bleeding and ensure complete tumor removal.¹² Recurrence is a known concern, with studies indicating that hemangiomatous ameloblastomas have a tendency for local recurrence, underscoring the need for long-term follow-up.¹³ Al-

though the overall prognosis is generally favorable, recurrence rates highlight the importance of adequate surgical margins and post-surgical monitoring.¹⁴

Only 24 cases have been reported in literature as of 2024 to the best of our knowledge. This is the 25th case being reported and it aims to provide a detailed account of this variant, contributing to a better understanding of its diagnostic challenges. The goal is to add to the existing literature, which is limited by the infrequency of such cases, and to emphasize the need for

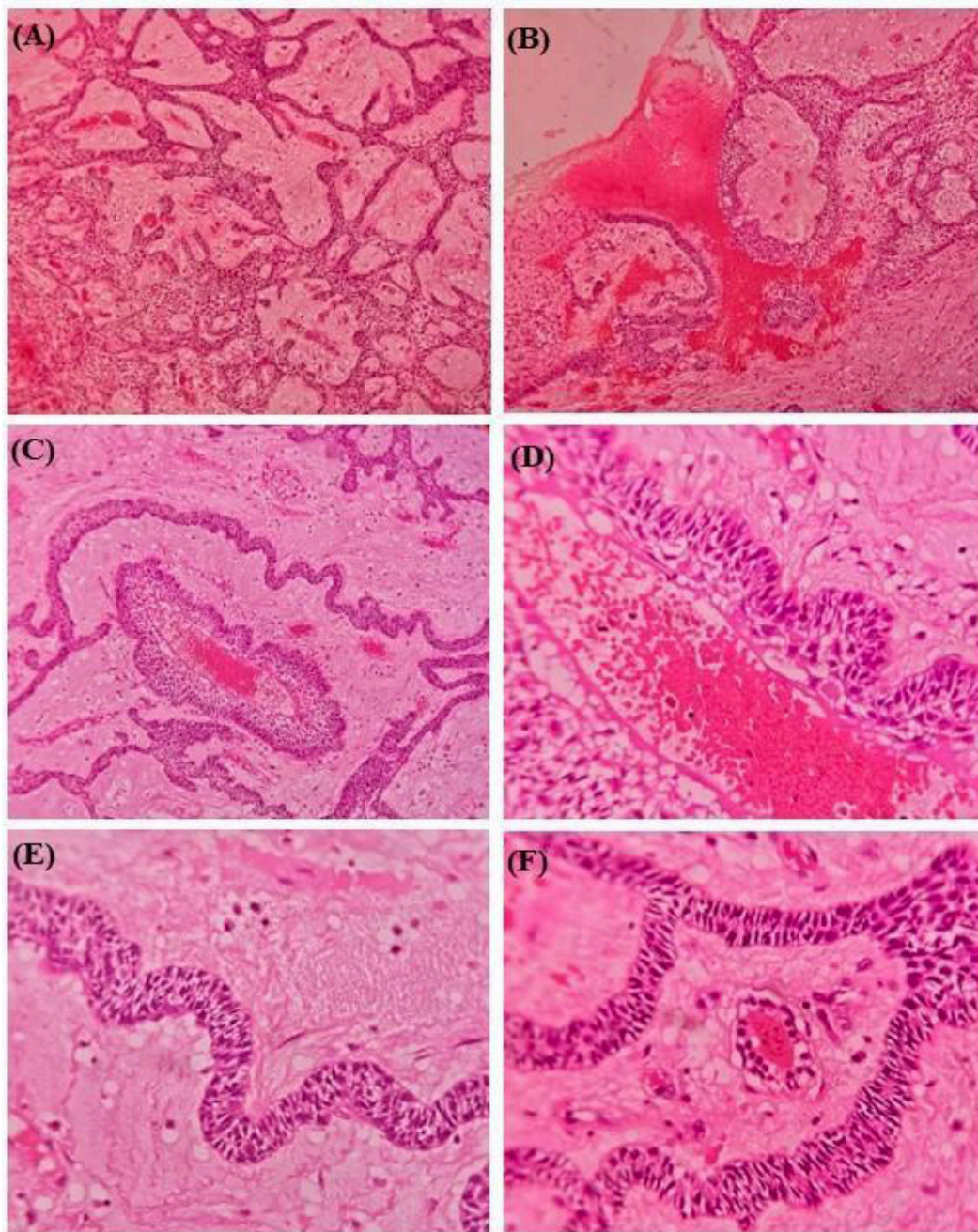


Fig. 1: numerous blood vessels present in the intervening connective tissue (A), large blood-filled spaces (B), blood filled space present within a large sized follicle (C), higher magnification of (C) is (D), Interconnecting strands lined by low cuboidal to tall columnar cells (E), endothelium lined blood vessel in the intervening stroma (F).

careful clinical and histological evaluation to ensure optimal patient outcomes.

CASE REPORT

A 21-year-old male reported to a private clinic with the chief complaint of swelling in relation to the right posterior mandible. The patient did not give a history of pain or any other symptoms. On radiographic examination, the OPG revealed a wide radiolucent lesion with sclerotic border seen extending from the body of the mandible up to the coronoid and condyle. Based on the clinical findings, ameloblastoma was given as the provisional diagnosis. Incisional biopsy of the lesion was done and paraffin embedded tissue blocks were sent to the department of oral and maxillofacial pathology in Saveetha dental college for further analysis.

H&E-stained sections were prepared and examined under a microscope. The sections revealed odontogenic epithelium arranged predominantly in the form of interconnecting strands and cords forming a plexiform pattern with areas of stromal degeneration. The epithelium was lined by peripheral low cuboidal to tall columnar ameloblast-like cells showing palisading of nucleus, reversal of polarity and sub nuclear vacuolation and central loose dis cohesive stellate reticulum like cells. Few follicles of varying size and shape with blood filled spaces were evident. The intervening dense connective tissue stroma showed intense vascularity with numerous endothelium lined blood vessels and large blood-filled spaces. The neoplastic odontogenic epithelium was also seen merging with the overlying surface epithelium suggestive of soft tissue extension of the lesion. Based on the histopathological findings, plexiform hemangiomatous ameloblastoma with soft tissue extension was given as the diagnosis.

DISCUSSION

Hemangiomatous ameloblastoma (HA) is a rare variant of ameloblastoma characterized by the presence of both ameloblastic epithelial islands and prominent vascular channels. The rarity of this variant has led to limited understanding and a scarcity of well-documented cases. This is the 25th case reported in literature all over the world and the 12th case reported in India. The clinical presentation of HA can be quite variable, often resulting in delayed diagnosis or misdiagnosis. While conventional ameloblastomas are typically slow-growing and asymptomatic, HA may present with more aggressive symptoms such as swelling, bleeding, or rapid expansion, due to the presence of vascular structures within the tumor.¹¹ The vascular component in HA has been described as resembling hemangiomas, which can make differentiation from other vascular lesions challenging.¹⁵

From the literature, it can be noted that the age range of the patients with HA ranged from 13-51 years with the mean age being 50 years with a slight predominance in males and a male to female ratio of 12:11. 88% of the cases occurred in the mandible similar to our case. The radiographic features of hemangiomatous ameloblastomas are often not distinct from those of conventional ameloblastomas, which include mixed radiolucent-radiopacity on panoramic radiographs and CT scans.¹⁶ However, the presence of vascular spaces can occasion-

ally influence the radiographic appearance, potentially leading to misinterpretation as a more common vascular lesion like a hemangioma or lymphangioma.¹⁷ For this reason, the definitive diagnosis of HA cannot rely solely on imaging and requires histopathological confirmation.

Histologically, hemangiomatous ameloblastoma shares features with both ameloblastoma and hemangioma, with vascular spaces being a distinctive feature. The vascular spaces can vary in size, and they may be filled with red blood cells or blood clots, further contributing to the unique presentation of this variant.⁴ Although this vascularity is a major component of HA, the epithelial structures still maintain the classic features of ameloblastoma, including the presence of peripheral columnar or cuboidal cells resembling the ameloblasts, often associated with a stellate reticulum-like central area.¹⁸ In the literature it can be seen that the most common variant of ameloblastoma accompanied with HA is the plexiform type. This mixed histological composition underscores the complexity of the tumor and necessitates a careful and thorough examination for accurate diagnosis.

Several theories have been formulated throughout the years describing the pathogenesis of these lesions. The first theory states that the induction of capillaries which give nutrition to the outer enamel epithelium results in formation of new blood vessels which later becomes a component of the tumor.¹⁹ Another theory suggests that the increase in vascular spaces is a result of traumatic injury caused during processes such as tooth extraction. Also, any disturbances during the formation of granulation tissue after injury can also lead to the formation of this tumor.²⁰ Multiple factors cause the angiogenesis process during tumour development.

Few authors also suggest that HA can be a collision type of tumor meaning when two types of tumors co-exist in the same area the tumor components collide and form a tumor with distinct features.²¹ Another theory suggests that the formation of blood filled spaces can be a secondary change where the blood vessels dilate and persist in cases where stromal cysts are present.²² The last theory says that the HA is not a distinct entity and that it is similar to the other types of ameloblastomas with variable amounts of vascularity.²³

Some of the key histopathological differential diagnoses of HA are vascular malformations or hemangiomas occurring in the oral cavity. These can present with significant vascular proliferation, but unlike hemangiomatous ameloblastoma, they do not exhibit the epithelial islands or follicles that are indicative of an ameloblastoma. The vascular pattern in hemangiomatous ameloblastoma is typically associated with solid tumor islands of ameloblastic cells, whereas true hemangiomas or vascular malformations are generally devoid of odontogenic epithelium. Telangiectatic osteosarcoma and angiomatoid malignant fibrous histiocytoma are also other differentials for HA.

Surgical resection remains the mainstay of treatment for hemangiomatous ameloblastomas, similar to other variants of ameloblastoma. However, due to the vascular nature of the tumor, special care is required during surgery to control bleeding, particularly in larger tumors.²⁴ Surgical approaches may involve complete enucleation, resection with a margin of



healthy tissue, or, in more aggressive cases, a segmental resection of the affected bone. The choice of surgical technique is influenced by factors such as tumor size, location, and the presence of recurrence.²⁵ Complete excision with adequate margins is essential to minimize the risk of recurrence, as incomplete removal can lead to a higher likelihood of local recurrence, a known characteristic of ameloblastomas in general this is very important factor to consider in health care.²⁶

Recurrence of hemangiomas ameloblastoma is a significant concern, and studies have shown that this variant exhibits a similar tendency for recurrence as conventional ameloblastomas.²⁷ The rate of recurrence can vary, but it is generally higher when tumors are incompletely excised, which underscores the importance of achieving clear surgical margins during the initial resection.²⁸ As of 2024 no recurrence has been reported in HA cases. Long-term follow-up is critical in cases of HA, as recurrence may occur several years after the initial surgery, requiring ongoing monitoring to detect any signs of regrowth.²⁹ In some cases, recurrent tumors may present with more severe clinical manifestations, making early detection and intervention crucial for preserving both function and aesthetics.

Despite the potential for recurrence, the overall prognosis for patients with hemangiomas ameloblastomas is generally favorable when treated adequately. These tumors are benign and typically do not metastasize, although the local invasion and aggressive growth patterns necessitate careful management.⁴ The presence of the vascular component does not appear to alter the biologic behavior significantly compared to conventional ameloblastomas, but it may complicate the surgical approach due to bleeding concerns.³⁰ Additionally, the rarity of HA and the limited number of case reports in the literature highlight the need for more research into the long-term outcomes and the best practices for managing this variant.

CONCLUSION

In conclusion, hemangiomas ameloblastoma represents a rare and unique form of ameloblastoma that poses diagnostic and therapeutic challenges. Its combination of ameloblastic and vascular elements necessitates careful histopathological examination and thoughtful consideration of surgical treatment options. Further research into the pathogenesis of HA is needed to better understand its biological behavior, as well as to establish more standardized treatment protocols. More studies on this rare variant will help the clinicians to be better equipped to recognize, diagnose, and manage hemangiomas ameloblastomas, ultimately improving patient outcomes, promote well being and reducing the risk of recurrence.

REFERENCES

- Lee SK, Kim YS. Current concepts and occurrence of epithelial odontogenic tumors: I. Ameloblastoma and adenomatoid odontogenic tumor. *Korean journal of pathology*. 2013;47.
- El-Naggar AK, Chan JKC, Grandis JR, Takata T, Slootweg PJ. WHO Classification of Head and Neck Tumours. Lyon: IARC Press; 2017.
- Kasangari MD, Gundamaraju K, Jyothsna M, Subash AV, Aravind K. Hemangiomas ameloblastoma-A case report of a very rare variant of ameloblastoma. *Journal of clinical and diagnostic research*. 2015;9(5).
- Sankar R, John S, Devi P, Verma S, Singh P, Gupta S. Hemangiomas ameloblastoma; A separate entity?'. In *Seminars in Diagnostic Pathology*. Vol. 42. WB Saunders; 2025.
- Childers EL, Taddasse-Heath L, Bonnicksen A, Naab T. Vascularized ameloblastoma: A case report and clinicopathologic review of 18 cases from the literature. *Oral Surgery, Oral Medicine, Oral Pathology and Oral Radiology*. 2020;129:e264-268.
- Da Cunha RG, Da Costa DLL, Cavalcanti TR, Libório-Kimura TN, Travassos RL, Castelo Branco ML, et al. Hemangiomas ameloblastoma of the mandible: A report of a rare case. *Oral Surg Oral Med Oral Pathol Oral Radiol*. 2018 Sept;126(3):e69. Available from: <http://dx.doi.org/10.1016/j.oooo.2018.02.175>
- Choudhary A, Sagar P, Prasad K, Ranganath K. Unicystic ameloblastoma masquerading as vascular lesion- A case report. *Research Square*. 2024. Available from: <http://dx.doi.org/10.21203/rs.3.rs-4523468/v1>
- Venigalla A, Bojji M, Piniseti S, Babburi S. Hemangiomas ameloblastoma: Case report with a brief review. *J Oral Maxillofac Pathol*. 2018;22(4):24. Available from: http://dx.doi.org/10.4103/jomfp.jomfp_28_16
- Choi WJ, Lee P, Thomas PC, Rath TJ, Mogensen MA, Dalley RW, et al. Imaging approach for jaw and maxillofacial bone tumors with updates from the 2022 World Health Organization classification. *World J Radiol*. 2024 Aug 28;16(8):294-316. Available from: <http://dx.doi.org/10.4329/wjr.v16.i8.294>
- Puri PD, Krishna A, Gosavi S, Nayyar V. Hemangiomas ameloblastoma with spindle cell proliferation: A rare case report and review of literature. *J Oral Maxillofac Pathol* 2022 Jan;26(1):132-3. Available from: http://dx.doi.org/10.4103/jomfp.jomfp_394_21
- Van Rensburg LJ, Thompson IO, Kruger HE, Norval EJ. Hemangiomas ameloblastoma: Clinical, radiologic, and pathologic features. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology*. 2001;91:374-80.
- Neagu D, Escuder-de la Torre O, Vázquez-Mahía I, Carral-Roura N, Rubín-Roger G, Penedo-Vázquez Á, et al. Surgical management of ameloblastoma. Review of literature. *J Clin Exp Dent*. 2019 Jan;11(1):e70-5. Available from: <http://dx.doi.org/10.4317/jced.55452>
- De Ac Almeida R, Andrade ED, Barbalho JC, Vajgel A, Vasconcelos BD. Recurrence rate following treatment for primary multicystic ameloblastoma: systematic review and meta-analysis. *International journal of oral and maxillofacial surgery*. 2016;45:359-67.
- Milman T, Ying GS, Pan W, Livolsi V. Ameloblastoma: 25 year experience at a single institution. *Head and neck pathology*. 2016;10:513-20.
- Routray S, Mishra S, Mohanty N, Panda S, Panda P. Recurrent aggressive multicystic ameloblastoma with hemangiomas proliferation and giant cells: A perplexed diagnosis. *Indian J Med Sci*. 2020 Feb 25;71(93):93-6. Available from: http://dx.doi.org/10.25259/ijms_15_19
- Kim SG, Jang HS. Ameloblastoma: a clinical, radiographic, and histopathologic analysis of 71 cases. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology*. 2001;91:649-53.
- Rajmohan M, Prasad H, Shanmugasundaram N, Thangam PT, Ilayaraja V, Anuthama K. Hemangiomas Ameloblastoma: A Rare Variant. *J Orofac Res*. 2014;4:63-6. Available from: <http://dx.doi.org/10.5005/jp-journals-10026-1130>
- Jois HS, Kumar K.P M, Kumar MS, Waghrey S. A mixed neoplasm of intraosseous hemangioma with an ameloblastoma: A case of collision tumor or a rare variant? *Clin Pract*. 2011 Dec 30;2(1):e5. Available from: <http://dx.doi.org/10.4081/cp.2012.e5>
- Sharma VK, Verma SK, Goyal L, Chaudhary PK. Hemangiomas ameloblastoma in maxilla: A report of a very rare case. *Dent Res J (Isfahan)*. 2012 May;9(3):345-9. Available from: <https://www.ncbi.nlm.nih.gov/pubmed/23087743>
- Tamgadge AP, Sirur D, Bhalarao S, Pereira T, Tamgadge S. Hemangiomas Ameloblastoma: A case report of a rare variant of ameloblastoma. *International Journal of Contemporary Dentistry*. 2010;1(2).
- Saxena P, Ahmed N, Mathur R. Follicular variant of



- ameloblastoma with cavernous hemangioma in maxilla: a rare example of collision tumor. *Int J Health Sci Res.* 2020;10(7):81–6.
22. Lucas RB. A vascular ameloblastoma. *Oral Surgery, Oral Medicine, Oral Pathology.* 1957;10:863–8.
 23. Smith JF. Report of thirty cases. *Oral Surgery, Oral Medicine, Oral Pathology.* 1960;13:1253–7.
 24. Dash KC, Ramachandra S, Bhuyan L, Kumar H. An insight of intramural plexiform hemangiomatous proliferation in unicystic ameloblastoma: A rare case report. *Journal of Dr NTR University of Health Sciences.* 2018 July;7(3):228–32. Available from: http://dx.doi.org/10.4103/jdrntruhs.jdrntruhs_58_17
 25. Surana KA, Pandiar D, Krishnan RP. Immunohistochemical expression of MDM2, bcl-2, SATB2 and ki-67 in histological variants of unicystic ameloblastoma. *Head Neck Pathol.* 2024 Oct 15;18(1). Available from: <http://dx.doi.org/10.1007/s12105-024-01705-7>
 26. Kumar VS, Kumar PR, Yadalam PK, Anegundi RV, Shrivastava D, Alfurhud AA, et al. Machine learning in the detection of dental cyst, tumor, and abscess lesions. *BMC Oral Health.* 2023 Nov 6;23(1). Available from: <http://dx.doi.org/10.1186/s12903-023-03571-1>
 27. Krishnan RP, Pandiar D, Sagar S. Immunohistochemical expression of CK14 and Bcl-2 in odontogenic keratocyst and its variants. *Appl Immunohistochem Mol Morphol.* 2024 Mar 1;32(3):151–6. Available from: <http://dx.doi.org/10.1097/PAI.0000000000001182>
 28. Gupta N, Kumar P, Malu M, Marandi M. Hemangiomatous ameloblastoma with dentinoid formation: A Sui generis case report. *International Journal of Medical and Oral Research.* 2023 Jan;8(1):35–9. Available from: http://dx.doi.org/10.4103/ijmo.ijmo_1_23
 29. Adisa AO, Lawal AO, Olusanya AA, Arotiba JT. Hemangiomatous ameloblastoma: report of a case. *Niger Dent J.* 2010 July 1;18(2):73–5. Available from: <http://dx.doi.org/10.61172/ndj.v18i2.146>
 30. Kasangari MD, Gundamaraju K, Jyothsna M, Subash AV, Aravind K. Hemangiomatous ameloblastoma- A case report of a very rare variant of ameloblastoma. *J Clin Diagn Res.* 2015 May;9(5):ZD08-10. Available from: <http://dx.doi.org/10.7860/JCDR/2015/13331.5886>

