

Hemangiomatous Ameloblastoma- A Case Report of a Very Rare Variant of Ameloblastoma

MARUTHI DEVI KASANGARI¹, KIRAN GUNDAMARAJU², MANDAPATI JYOTHSNA³,
ANDY VENKATA SUBASH⁴, KUMBAKONAM ARAVIND⁵

ABSTRACT

Ameloblastoma is a true benign odontogenic neoplasm with many classical histological variants, common being follicular and plexiform types. Hemangiomatous ameloblastoma is a very rare variant that shows unique histopathologic characteristics varying from conventional ameloblastoma. We present a rare variant of ameloblastoma in a 35-year-old female patient with a swelling over left mandibular region, showing mixed radiolucent-opacity, which on enucleation histopathologically revealed ameloblastomatous areas with extensive vascular component.

Keywords: Collision tumour, Follicular ameloblastoma, Haemangioma, Odontogenic tumour, Plexiform ameloblastoma

CASE REPORT

A 35-year-old female patient presented with a chief complaint of painless swelling on the left side of the face since eight months to the Department of Oral Medicine and Radiology, Malla Reddy Dental College for Women, Hyderabad. Telangana, India. Her history revealed that the swelling gradually grew to the present size. Her medical, dental, habitual and family history was not significant. On extra oral examination, inspection showed a swelling measuring approximately 4x3 cm in size, oval in shape, having well defined margins. The colour of the lesion was similar to the normal adjacent skin. The swelling extended superiorly to the line extending from the tragus of the ear to the corner of the mouth. Inferiorly to lower border of the mandible, posteriorly up to the ramus of the mandible and anteriorly up to the corner of the mouth [Table/Fig-1]. No visible pus discharge was seen. On palpation, the inspeitory findings were confirmed and the lesion was bony hard with central region showing variable consistency of hard and soft areas. The lesion was non tender. On intra oral examination, firm, smooth, non-fluctuant swelling was seen extending from 31 to the anterior border of the ramus. The mandibular left first molar and second premolar have been extracted 11 months back. First premolar was carious.

Panoramic radiograph revealed an well defined mixed radioopaque-lucent multilocular lesion extending from lower left central incisor to the ascending border of ramus [Table/Fig-2]. Impacted second molar was seen at the inferior border of the mandible. Few septae were arranged in tennis racket appearance. Root resorption was noticed in relation to 34. Over retained 85 was also seen. Radiological

differential diagnosis of central hemangioma, ameloblastoma, odontogenic myxoma was considered.

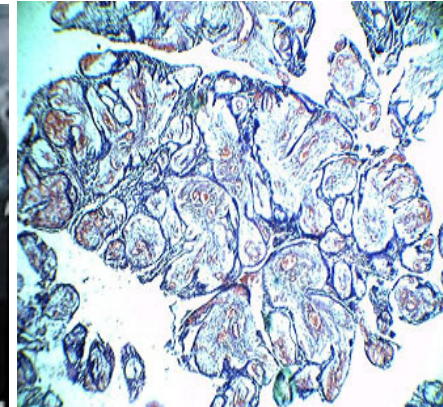
A provisional diagnosis of ameloblastoma and clinical differential diagnosis of dentigerous cyst and odontogenic keratocyst was made. After obtaining patient's consent, enucleation was done and multiple bits of the soft tissue specimen of approximately 4cm X 2cm X 1cm were sent to the Department of Oral Pathology.

On microscopic examination, the tissue section revealed plexiform ameloblastoma with a prominent vascular component. It showed anastomosing cords and sheets of odontogenic epithelium in a loosely arranged vascular connective tissue stroma. The epithelium was surrounded by cuboidal ameloblast like cells with central stellate reticulum like cells. The vascular component consisted of blood filled spaces of varying sizes lined by endothelial cells. At focal areas vascular spaces were large and compressing the odontogenic strands [Table/Fig-3-5]. A diagnosis of Hemangiomatous Ameloblastoma was made.

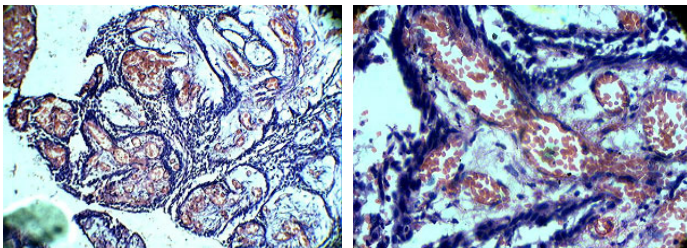
Partial hemimandibulectomy was planned followed by immediate reconstruction with a nonvascularised iliac graft. But the patient was not cooperative and reluctant for surgical treatment and hence the procedure could not be carried out. The patient was recalled for follow up visits but did not return to the department.

DISCUSSION

Ameloblastoma is one of the most common odontogenic tumours with approximately making 11% of them. It is categorized based



[Table/Fig-1]: Extra oral view showing swelling [Table/Fig-2]: OPG showing mixed radiolucent-opacity [Table/Fig-3]: Photomicrograph (5x) showing odontogenic cords and islands with loose vascular tissue



[Table/Fig-4]: Photomicrograph (10x) showing ameloblastomatous areas with vascular areas **[Table/Fig-5]:** Photomicrograph (40x) showing extensive vascular area

on clinico-radiological features into three types: solid or multicystic, unicystic, and peripheral [1,2]. Odontogenesis is a complex process and any deviation from normal process results in odontogenic cysts and tumours. In ameloblastoma there is faulty differentiation of enamel organ to point of actual enamel formation [2,3].

Various histological variants of ameloblastoma have been described in the literature like follicular (most common), plexiform, acanthomatous, granular (most aggressive), basal cell and desmoplastic [4]. Ameloblastoma has many histological variants with follicular ameloblastoma the most common one. Reports of variants like clear cell, papilliferous, keratoameloblastoma and a very rare pattern hemangiomatous ameloblastoma have also been described in the literature. Studies have shown that all the mentioned histological variants do not differ significantly in either biological behaviour of the tumour or its prognosis after treatment. Desmoplastic and hemangiomatous ameloblastoma are exceptions with the former thought to be less aggressive and the later due to its extreme vascular component carry surgical complications. However, in Many cases have been reported with mixture of these patterns [4-6].

Hemangiomatous ameloblastoma is a very rare variant of ameloblastoma. Hemangiomatous ameloblastoma term refers to any ameloblastoma which has in its stroma many spaces filled with blood or large endothelial-lined capillaries. First case of Hemangiomatous Ameloblastoma (vascular ameloblastoma) was described by Kuhn (1932) as a combination of hemangioma and adamantinoma. Aisenberg (1950) later reported a similar lesion as adamantinohemangioma. It is supposed that in Hemangiomatous Ameloblastoma the stromal component is mostly replaced by vascular component [7].

Earlier hemangiomatous ameloblastoma was documented as ameloblastic hemangiomas, adamantino hemangiomas and hemangio ameloblastomas [5,6]. Literature search to the best of our knowledge has revealed less than 15 cases of hemangiomatous ameloblastoma, suggesting its rarity [Table/Fig-6] [1,6-12].

As very few cases of hemangiomatous ameloblastoma are reported, the origin of its vascular component is still debatable and not clearly understood. Few authors consider the vascularity as hamartomatous growth, few others as part of neoplastic process and others consider it as a separate neoplasm. Whereas Smith did not considered hemangiomatous ameloblastoma as a separate variant of ameloblastoma as according to him the extensive vascularity might be owed to other causes [8].

To summarize the various theories proposed to explain the pathogenesis of the vascular component of hemangiomatous ameloblastoma are [6-11,13]

1. Angiogenesis during tumour development.
2. Represents a separate neoplasm,
3. Hamartomatous malformation,
4. Traumatic incident,
5. Secondary change,
6. HA representing a collision tumour.

Sometimes during odontogenesis, anomalous induction of blood

Author	Age	Sex	Site	Treatment done	Healing and prognosis
Aisenberg MS [6]	48	female	Right posterior mandible	Enucleation	uneventful
Lucas RB [9]	43	female	Right mandible	Resection of affected portion	Uneventful and no complaint after follow up
van Rensburg LJ [10]	26	female	Left mandible, 3rd molar region	Partial hemimandibulectomy Planned but patient refused	No follow up could be done
Ide F [2]	56	male	Anterior maxilla	Enucleation and curettage	Good healing after 6 months follow-up
Tamgadge AP [1]	31	male	Left mandible premolar molar area	Enucleation and curettage	Good healing after 4 months follow-up
Sharma VK [11]	15	male	Maxillary right molar area	Enucleated	Followed for 6 months
Harshvardhan SJ [12]	42	male	Right posterior mandible	Hemimandibulectomy	Uneventful and no complaint after followup for 2 years
Sarode GS [13]	18	male	Right posterior mandible	curettage	Patient lost to follow-up
Rajmohan M [14]	20	male	Right posterior mandible	Hemimandibulectomy	Patient lost to follow-up
Our Case	35	Female	Left posterior mandible	Enucleation	Lost to follow up

[Table/Fig-6]: Comparison of various cases of Hemangiomatous Ameloblastoma

vessels may occur and result in their abnormal proliferation. Such proliferated vessels possibly will turn into a component of tumour [11].

Trauma during procedures like extraction of teeth might induce the epithelial cell rests in the periodontal ligament to proliferate and result in tumour process. Usually after any injury the injured tissues undergo repair to retain their original condition. Any disturbance in repair process of damaged tissues involves excessive granulation tissue formation, which consists of proliferating endothelial cells and new capillaries [13]. The presence of extensive vascular component in our case might be due to stimulation and proliferation of blood vessels due to trauma created during extraction of teeth and subsequent repair phenomenon.

Lesions such as hemangiomas, telangiectatic osteosarcoma, angiomatoid malignant fibrous histiocytoma should be included in differential diagnosis. In telangiectatic osteosarcoma, there are large spaces filled with blood and huge areas of necrosis. Whereas, angiomatoid malignant fibrous histiocytoma contain characteristics of a fibrohistiocytic tumour and a vascular tumour [7]. Surgical complications due to extensive vascularity in such vascular lesions should always be kept in mind during treatment planning.

Pathogenesis, clinical features, treatment modality and prognosis of hemangiomatous ameloblastoma until now is not completely understood as only very few cases have been reported. Few authors considered Hemangiomatous Ameloblastoma as a collision type of tumour, in which two separate tumours develop in the same area and collide, and their components blend together [13]. Lucas, viewed that vascular component in hemangiomatous ameloblastoma is purely a secondary change. According to him cystic degeneration occurs in stroma in plexiform ameloblastoma, and during the cysts formation some blood vessels instead of diminishing may persist and dilate resulting in vascular component as seen in hemangiomatous ameloblastoma [12,15].

CONCLUSION

Hemangiomas of Ameloblastoma and other such lesions with extensive vascular component may lead to fatality even following minor procedures. Our case clearly demonstrates the distinctive histologic pattern and should be kept in differential diagnosis of vascular lesions. More number of such cases has to be studied thoroughly to exactly know the origin, nature and behaviour of Hemangiomas of Ameloblastoma.

REFERENCES

- [1] Tamgadge AP, Sirur DG, Bhalerao SS, Pariera T, Tamgadge SA. Hemangiomas of Ameloblastoma: A case report of a rare variant of ameloblastoma. *Int J Contemp Dent*. 2010;2:16-19.
- [2] Ide F, Horie N, Shimoyama T, Sakashita H. So-called hybrid odontogenic tumours: Do they really exist. *Oral Med Pathol*. 2001;6:13-21.
- [3] Shafer WG, Hine MK, Levy BM. Cysts and tumours of odontogenic origin. *Shafer's Textbook of Oral Pathology* (5th ed). New Delhi: Elsevier 2005; 395-98.
- [4] Krammer IR, Pindborg JJ, Shear M. Histological typing of odontogenic Tumours. Berlin: Springer-Verlag; 1992. pp. 11-14.
- [5] Neville BW, Damm DD, Allen CM, Bouquot JE. Oral and maxillofacial Pathology. 2nd Ed., Philadelphia. WB Saunders Co.; 2002. pp. 611-16.
- [6] Aisenberg MS. Adamantinoid hemangioma. *Oral Surg Oral Med Oral Pathol*. 1950;3:798-801.
- [7] Oliver RT, McKenna WF, Shafer WY. Hemangiomas of ameloblastoma; report of a case. *J Oral Surg Anesth Hosp Dent Serv*. 1961;19:245-48.
- [8] Smith JF. The controversial Ameloblastoma. *Oral Surg Oral Med Oral Pathol*. 1968;26:45-75.
- [9] Lucas RB. A vascular ameloblastoma. *Oral Surg Oral Med Oral Pathol*. 1957;10(8):863-68.
- [10] Van Rensburg LJ, Thompson IO, Kruger HE, Norval EJ. Hemangiomas of ameloblastoma: Clinical, radiologic, and pathologic features. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2001;91:374-80.
- [11] Sharma VK, Verma SK, Goyal L, Chaudhary PK. Hemangiomas of ameloblastoma in maxilla: A report of a very rare case. *Dent Res J*. 2012;9:345-49.
- [12] Harshvardhan SJ, Mohan Kumar KP, Kumar MS, Waghrey S. A mixed neoplasm of intraosseous hemangioma with an ameloblastoma: A case of collision tumour or a rare variant. *Clinics and practice*. 2012;2(1):e5.
- [13] Sarode GS, Sarode SC, Vaidya K. Intraluminal plexiform hemangiomas of ameloblastomatous proliferation in unicystic ameloblastoma: An unusual case report. *Indian J Dent Res*. 2013;24:390-92.
- [14] Rajmohan M, et al. Hemangiomas of ameloblastoma: A Rare Variant. *J Orofac Res*. 2014;4(1):63-66.
- [15] Villa VG. Hemangioma of the cavernous type associated with ameloblastoma (ameloblastic hemangioma). *J Oral Surg Anesth Hosp Dent Serv*. 1960;18:429-31.

PARTICULARS OF CONTRIBUTORS:

1. Reader, Department of Oral & Maxillofacial Pathology, Malla Reddy Dental College for Women, Hyderabad, Telangana, India.
2. Assistant Professor, Department of Oral & Maxillofacial Pathology, Government Dental College and Hospital, Hyderabad, Telangana, India.
3. Associate Professor, Department of Oral & Maxillofacial Pathology, Government Dental College and Hospital, Hyderabad, Telangana, India.
4. Reader, Department of Oral & Maxillofacial Pathology, Lenora Dental College, Rajamundry, Andhra Pradesh, India.
5. Senior Lecturer, Department of Oral Medicine & Radiology, Malla Reddy Dental College for Women, Hyderabad, Telangana, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:

Dr. G Kiran,
Assistant Professor, Department of Oral & Maxillofacial Pathology, Government Dental College & Hospital,
Afzalgunj, Hyderabad-500012, Andhra Pradesh, India.
E-mail : Kiran.dentist@gmail.com

Date of Submission: **Feb 04, 2015**
Date of Peer Review: **Mar 15, 2015**
Date of Acceptance: **Mar 30, 2015**
Date of Publishing: **May 01, 2015**

FINANCIAL OR OTHER COMPETING INTERESTS: None.