International Journal of Health Sciences and Research
Vol.10; Issue: 7; July 2020
Website: www.ijhsr.org
Case Report *ISSN: 2249-9571*

Follicular Variant of Ameloblastoma with Cavernous Hemangioma in Maxilla: A Rare Example of Collision Tumor

Pallavi Saxena[1], Naiem Ahmed[2], Ranjan Mathur[3]

[1,2]Senior Resident, [3]Professor and Head,
Department of Dentistry, Sardar Patel Medical College and Associated Hospitals, Bikaner, INDIA

Corresponding Author: Pallavi Saxena

ABSTRACT

Collision tumors are those which are of two distinct histological features and tend to occur at same anatomical site. One such rare collision tumor is a hemangiomatous ameloblastoma. This tumor is unique as two neoplasms one that of vascular origin and another of odontogenic origin tend to occur at one site. But due to paucity of reported cases the exact mechanism of formation is not known. Therefore, we report a case of such rare entity. A 32 year female patient reported to the outpatient department of our institute. She presented with a massive growth present in anterior maxilla region present since four months. She underwent an excisional biopsy which came out to be reported as Hemangiomatous Ameloblastoma. The healing was uneventful. Hemangiomatous Ameloblastoma is considered as a variant of solid multicystic ameloblastoma. On a thorough literature search we found that till date only eleven cases have been reported. Due to this scarcity of reported cases, the exact pathogenesis is less understood. Moreover the clinical behaviour is also less studied which makes it important for this neoplasm to be considered as a separate entity.

Keywords: Hemangiomatous ameloblastoma, Collision tumor, Follicular Ameloblastoma, Cavernous Hemangioma.

INTRODUCTION

Hemangioma is a benign neoplasm of dilated blood vessels. These appear within few weeks of birth. Hemangiomas are characterized by the hyperplasia of veins and capillaries.[1] Different hypothesis describe hemangiomas differently, some called them reactive while a few have stated them as a purely neoplastic process. Hemangiomas were first described by Mulliken and Glowaki in the year 1982, giving a biological classification. Later in the year they were classified as capillary and cavernous hemangioma by Shafer et al in the year 1993.[2] Histologically, it is a tumor of mesenchymal origin and involves proliferation of endothelial cells lining the vascular blood vessels. But the scope of this article not only involves hemangiomas rather a rare presentation of hemangioma with ameloblastoma (AB).

The other component of this case was AB which are neoplasms of odontogenic origin. AB are second most common among the odontogenic tumors. Clinically, AB is subdivided into solid multicystic, unicystic and peripheral type. Histologically, it has two most common variants which are the follicular and plexiform.[3] Other histological variants known to occur are the granular cell and acanthomatous type. While few very rarely seen are the basal cell, desmoplastic and clear cell types.[4] A very unusual variant of is AB having histological features of Hemangioma. This biological feature of AB is of questionable pathogenesis. It is called as Hemangiomatous Ameloblastoma (HA).

It is a very rare variant of AB and on thorough literature search we found that till date only eleven cases have been reported so far. Therefore, we are reporting a case on HA, along with review of literature and discussion on possible pathogenesis of such uncertain presentation of AB.

CASE REPORT

A 32 year female patient presented to the outpatient department of our institute. She had a complain of a growth present on the right side of upper jaw which was disfiguring her face and was present since four months. On clinical examination of the patient, a growth measuring about 4.0x5.0 cm^2 was found which was firm in consistency, with normal overlying mucosa. It was obliterating the buccal vestibule and there was mobility in 13, 14 and 15. Extra orally the growth was causing the disfigurement of the right side of face involving the right cheek and malar region. Examination of the nasal cavity showed that the right side of the nasal chamber was obliterated with the growth and when asked the patient, it was found that she faced difficulty in breathing from the right nostril.

Fig 1: Extra-oral examination revealed a diffuse swelling causing disfigurement of face on the right side.

A Paranasal sinus view (Water's position) was advised along with a 3D CT-scan of the face and maxillary sinus. The PNS view showed a mixed radiopaque-radiolucent lesion with well defined borders involving the right maxilla, obliterating the right maxillary sinus, the right nasal cavity and the right orbital cavity. On the other hand, the lateral section of face and maxillary sinus on a 3D CT-scan showed a radio dense lesion involving the maxillary sinus, nasal cavity, orbital cavity and approaching the base of skull. Looking at the extensive size of the tumor and locally aggressive nature, a provisional diagnosis of AB was given. Furthermore, an aspiration was attempted but there was no yield which proved the solid nature of the tumor.

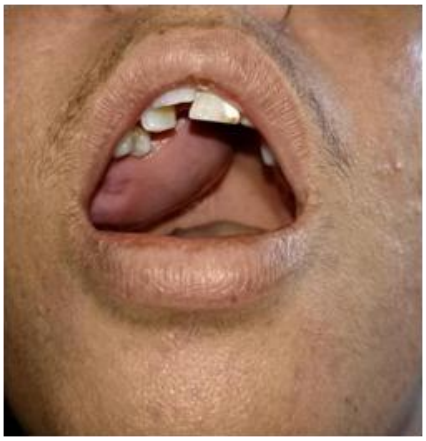

Fig 2: Intra-oral examination revealed a mass involving the anterior palate along with mobility in 13, 14 and 15.

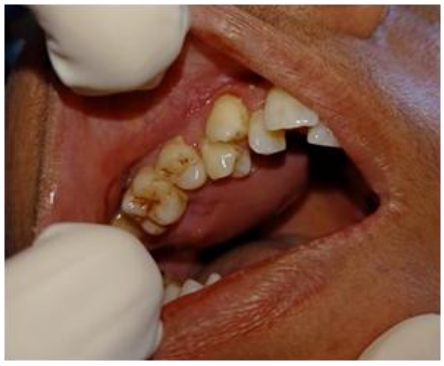

Fig 3: The illustrated image shows obliteration of the buccal vestibule

An excisional biopsy was performed, after obtaining patient's consent. There multiple excised bits and greyish brown in color with irregular borders and measuring around 4.0x5.0 cm to 2.0x3.0 cm. These specimens were sent to the pathology department for histopathological evaluation. The Hematoxylin and eosin stained sections showed two distinct types of histopathological features. One of those were discrete islands of tumor which were composed of peripheral layer of tall

columnar to cuboidal cells having polarized nuclei which were suggestive of ameloblast like cells. These tall cells enclosed the loosely arranged polyhedral cells resembling the stellate reticulum. These features were suggestive of follicular ameloblastoma. The other histopathological component was presence of large blood filled sinuses which were lined by endothelial cells. There was a remarkable proliferation of ovoid to plump endothelial cells. The surrounding stroma was variable in density. These features were suggestive of a cavernous hemangioma. Based on these collaborative histological features a final diagnosis of hemangiomatous variant of ameloblastoma was given.

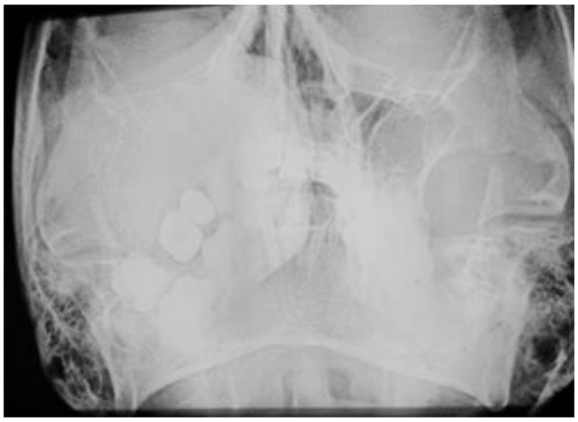

Fig 4: A paranasal sinus view (Water's Position) shows a mixed radiolucent-radioopaque lesion with well defined margins involving the right maxillary sinus, right nasal and orbital cavity.

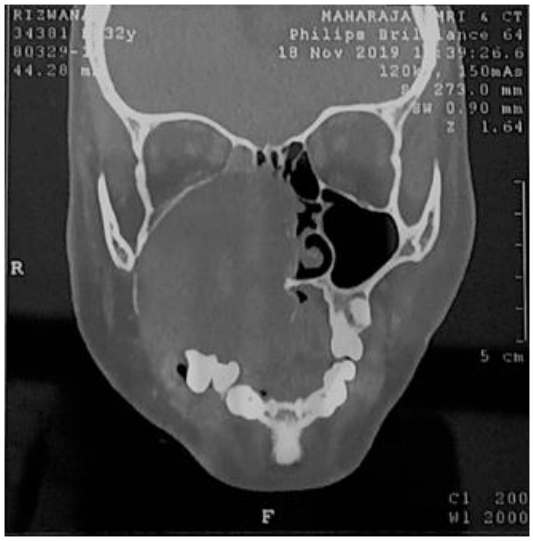

Fig 5: Transverse section of face and maxillary sinus shows a growth that involves right maxilla, maxillary sinus, nasal cavity and orbital cavity

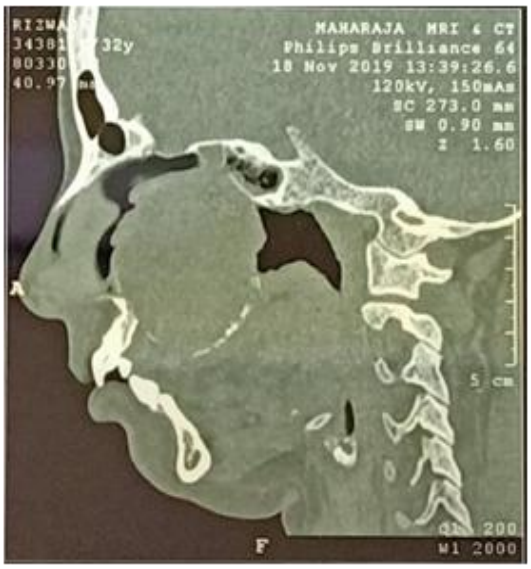

Fig 6: The CT scan on sagittal section showed the mass was extending posteriorly till the base of skull.

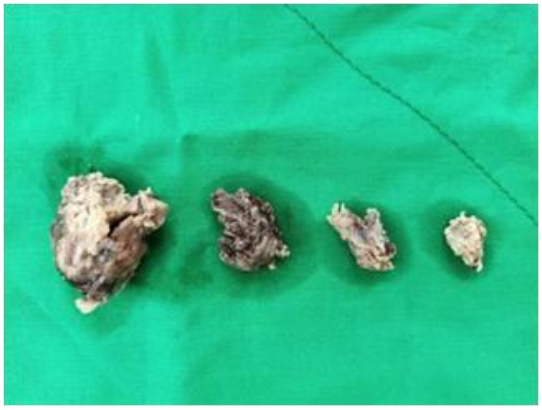

Fig 7: The gross pathology showed multiple excised bits and greyish brown in color with irregular borders and measuring around 4.0x5.0 cm to 2.0x3.0 cm.

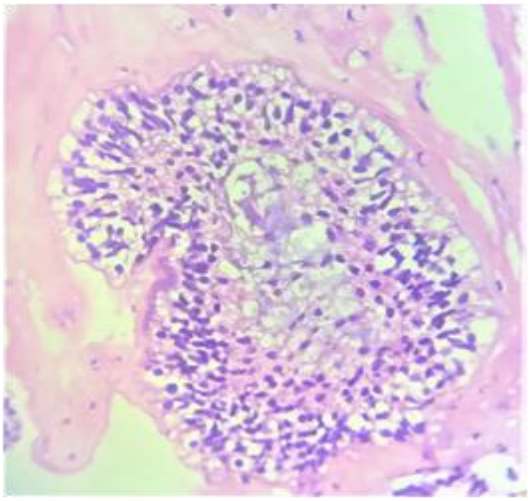

Fig 8: Hematoxylin and eosin stained sections showed islands of tumor which were composed of peripheral layer of tall columnar to cuboidal cells having polarized nuclei which were suggestive of ameloblast like cells. These tall cells enclosed the loosely arranged polyhedral cells resembling the stellate reticulum. These features were suggestive of follicular ameloblastoma.

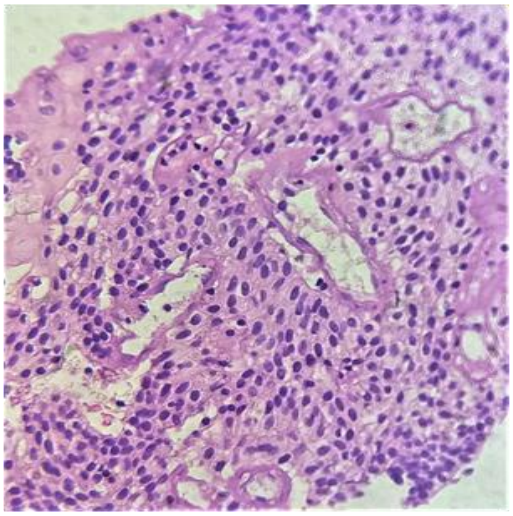

Fig 9: The H&E stained section under high magnification shows proliferation of round to ovoid plump cells suggestive of endothelial cells.

DISCUSSION

Hemangiomatous variant Ameloblastoma was first described by Kühn in 1932, where he described this variant as a type of solid multicystic ameloblastoma, in which a part of tumor consists spaces filled with blood or enlarged endothelial lined blood capillaries.[5] On a thorough literature search, we found that since the first description only eleven cases have reported so far.[6-16] This shows the infrequency of this pathological entity. Furthermore, it is because of the rarity in the frequency of this lesion the exact pathogenesis has not yet been found.

Table 1: This table shows comparison of previously reported cases of hemangiomatous ameloblastoma.

SN	Previous Cases	Age/ Gender	Site	Radiological Features	Possible Etiological Factors	Type of Biopsy	Prognosis
1.	Aisenberg6 (1950)	48/F	Posterior Mandible	-	-	Enucleation	Uneventful
2.	Lucas7 (1957)	43/F	Right Side of Mandible	-	-	Resection of the affected part	-
3.	van Rensburg8 (2001)	26/F	3rd molar region of Mandible left side	-	-	Partial Hemimandibulectomy planned, but the patient refused.	-
4.	Ide et al9 (2001)	56/M	Anterior Maxilla	-	-	Enucleation and Curettage	Uneventful healing
5.	Avinash et al10(2010)	31/M	Body of Mandible region left side	Well defined radiolucency	-	Hemimandibulectomy	-
6.	Jois et al 11(2012)	42/M	Posterior Mandible Right side	Mixed radiolucent radioopaque lesion	-	Hemimandibulectomy	-
7.	Sharma et al12(2012)	15/M	Maxilla Right side	Well defined radiolucency with sclerotic borders	-	Enucleation	-
8.	Sarode13 (2013)	18/M	Right side of Mandible	Well circumscribed multilocular radiolucent lesion	Tooth extraction	Curettage	-
9.	Rajmohan14 (2014)	20/M	Right Side of Mandible	Well defined multilocularity	Tooth extraction	Hemimandibulectomy	-
10.	Kasangari et al15 (2015)	35/M	Posterior Mandible Left Side	Well defined mixed radiolucent-radioopaque lesion	Tooth extraction	Enucleation	-
11.	Venigalla et al16(2016)	35/F	Posterior Mandible Right Side	Mixed Lesion	Trauma	Hemimandibulectomy	-
12.	Present Case (2020)	32/F	Anterior Maxilla	Mixed Radioopaque-radiolucent lesion	Needle core Biopsy	Enucleation	Good Healing

The previously reported cases show that there is a male predominance.[9-15] Unlikely in our case the patient was a female. The age of occurrence in previous cases, shows a wide age range from 18-56 years.[17] In our case, the age of the patient was in accordance with this age range our patient was of 32 years. The site of occurrence in previous cases were majorly, posterior mandible.[9,12] Only two previous cases show the lesion site in accordance of our case, which was anterior maxilla. The former cases have all shown a mixed radiopaque-radiolucent lesion.[10-14] This

radiological feature was in compatibility of our case. The etiological factor thought for the development of vascular component in the previous cases, was a pre-existing history of either trauma or tooth extraction.[13-16] On the contrary, in our case the patient had a history of needle core biopsy, from some other dental facility during the initial growth phase of the tumor. There are several proposed mechanisms about the angiogenesis in this tumor. But the exact theory is yet to be found out.

The various possible reasons for angiogenesis in an ameloblastoma are postulated by many researchers.[17,18,19] One of the theory states that the vascular component arises as a hamartomatous growth in ameloblastoma.[17] On the other hand, few authors consider the hemangiomatous growth as a separate neoplastic process and later on these two different entities collision with each other forming a collision tumor.[18] It is also stated postulated that, the stroma of ameloblastoma is of myxoid nature and sometimes due to degeneration of the stroma the blood vessels which are left behind can sometimes rupture and lead to accumulation of blood in connective tissue spaces.[11]

It is also postulated that during the process of odontogenesis, due to the inductive mechanism there can be excessive angiogenic phenomena.[17] Few authors have stated that at the time of tissue damage, the healing process takes place by the formation of granulation tissue which includes proliferation of the blood vessels, but sometimes, because of the neoplastic pathogenesis going on it may itself turn into another neoplasia. Another school of thought states trauma as one of the possible etiology. This could be due to extraction of tooth in area adjacent to tumor or physical injury to the site of tumor.[16] In our case, there was a history previously attempted needle core biopsy. Therefore, the theory of trauma could be the most accepted one.

So in conclusion, HA is a very rare phenomenon and due to scarcity of the reported cases, the exact pathogenesis is still questionable. Moreover, since there is a lot of process of genesis which happens in close vicinity of an aggressive tumor as that of AB, hence this collision tumor should be considered as separate entity.

REFERENCES

1. Waner M, Suen JY. A classification of congenital vascular lesions. In: Waner M, Suen JY, editors. Hemangiomas and Vascular Malformations of the Head and Neck. San Francisco: Wiley; 1999. p. 1-12.
2. Mulliken JB, Glowacki J. Hemangiomas and vascular malformations in infants and children: A classification based on endothelial characteristics. Plast Reconstr Surg 1982;69:412-22.
3. Rajendran R, Sivapathasundharam B, editors. Shafer's Textbook of Oral Pathology. 6th ed. New Delhi: Elsevier; 2009; 271-279.
4. Neville BW, Damn DD, Allen CM, Bouquot JE, editors. Oral and Maxillofacial Pathology. 3rd ed. Philadelphia: WB Saunders Co. 2009;702-707.
5. Reichart PA, Philipsen HP. Odontogenic tumors and allied lesions. London: Quintessence Publishing Co Ltd 2004;52-53.
6. Aisenberg MS. Adamantinohemangioma. Oral Surg Oral Med Oral Pathol. 1950;3: 798–801.
7. Lucas RB. A vascular ameloblastoma. Oral Surg Oral Med Oral Pathol. 1957;10:863–8.
8. van Rensburg LJ, Thompson IO, Kruger HE, Norval EJ. Hemangiomatous ameloblastoma: Clinical, radiologic, and pathologic features. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2001;91: 374–80.
9. Ide F, Horie N, Shimoyama T, Sakashita H. So-called hybrid odontogenic tumours: Do they really exists. Oral Med Patol. 2001; 6:13–21.
10. Avinash PT, Dhirendra GS, Sudhir SB, Pariera T, Sandhya AT. Hemangiomatous ameloblastoma: A case report of a rare variant of ameloblastoma. Int J Contemp Dent. 2010;1:16–9.
11. 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. 2012;2:e5.
12. Sharma VK, Verma SK, Goyal L, Chaudhary PK. Hemangiomatous ameloblastoma in maxilla: A report of a very rare case. Dent Res J (Isfahan) 2012; 9:345–9.
13. Sarode GS, Sarode SC, Vaidya K. Intraluminal plexiform hemangioamelo-blastomatous proliferation in unicystic ameloblastoma: An unusual case report. Indian J Dent Res. 2013;24:390–2.
14. Rajmohan M, Prasad H, Shanmugasundaram N, Tamil Thangam P, Ilayaraja V, Anuthama K. Hemangiomatous ameloblastoma: A rare variant. J Orofac Res. 2014;4:63–6.
15. 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;9: ZD08–10.
16. Venigalla A, Bojji M, Pinisetti S and Babburi S.Hemangiomatous Ameloblastoma: Case Report With a Brief Review. J Oral Maxillofac Pathol 2018 Jan;22(Suppl 1):S24-S28.
17. Villa VG. Hemangioma of the cavernous type associated with ameloblastoma (ameloblastic hemangioma). *J Oral Surg Anesth Hosp Dent Serv*. 1960;18:429-31.
18. Tamgadge AP, Sirur DG, Bhalerao SS, Pariera T, Tamgadge SA. Hemangiomatous Ameloblastoma: A case report of a rare variant of ameloblastoma. Int J Contemp Dent. 2010;2:16–9.

How to cite this article: 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):81-86.

******