Journal of Advanced Medical and Dental Sciences Research

@Society of Scientific Research and Studies ***NLM ID:*** *101716117*

Journal home page: www.jamdsr.comdoi: 10.21276/jamdsr Indian Citation Index (ICI) Index Copernicus value = 100

(e) ISSN Online: 2321-9599; (p) ISSN Print: 2348-6805

Case Report

Multitudinous presentation of mandibular cavernous hemangioma-A rare case report

[1]Rashmi Sathe, [2]Preeti Nair, [3]Akshay Agarwal, [4]Richa Bajpai

[1]Reader, [2]Professor and Head, [4]PG Resident, Department of Oral Medicine and Radiology, People's College of Dental Science and Research Centre, People University, Bhopal, Madhya Pradesh, India;
[3]Assistant Professor, Department of Oral Medicine and Radiology/People's Dental Academy, People University, Bhopal, Madhya Pradesh, India

ABSTRACT:
Introduction: Hemangiomasare hamartomatous proliferations of blood vessels. They are classified into Central, Capillary, and cavernous types. Hemangiomas rarely occur in the jaw, more commonly affecting the skull and vertebrae. Central hemangioma scan mimic other conditions due to their varied radiographic appearances.
A 54-year-old female presented with a 12-month history of swelling on the right side of her face. Examination revealed facial asymmetry, pain, and a bony hard swelling in the mandible. Radiographs showed a mixed radiopaque-radiolucent lesion with multilocular appearance. Provisional diagnoses included odontogenicmyxoma and ameloblastoma, while biopsy confirmed cavernous hemangioma. The lesion was excised enbloc, and reconstruction was done. Histopathology confirmed intraosseous cavernous hemangioma. **Conclusion:** Cavernous hemangiomas of the mandible are rare and present myriad radiographic features. Accurate diagnosis and multidisciplinary management are crucial to prevent complications and ensure effective treatment.
Keywords-Cavernous Hemangioma, honey comb,multilocular.

Received: 16 June, 2024 Accepted: 18 July, 2024

Corresponding author: Rashmi Sathe, Reader, Department of Oral Medicine and Radiology, People's College of Dental Science and Research Centre, People University, Bhopal, Madhya Pradesh, India

This article may be cited as: Sathe R, Nair P, Agarwal A, Bajpai R. Multitudinous presentation of mandibular cavernous hemangioma-A rare case report. J Adv Med Dent Scie Res 2024;12(8):84-88.

INTRODUCTION

Hemangiomas are abnormal proliferation of blood vessels,confused neoplasm and hamartoma. They are classified as Central, Capillaryand Cavernous.[1]These lesions present varied radiographic features are mostlymultilocular.[2]

SL Moore *et al*, [3] has mentioned maxilla to be commoner,while according toRP Langlais*et al*., mandibular site is commoner.[4]Higher predilection seen in females in 1st-3rd decade of life. Clinical features include pain, paraesthesia,expansionof the buccal and lingual cortex,tooth mobility.[5, 6]

The present case represents a rare Cavernous hemangioma of the right mandible with elusive radiographic features.

CASE REPORT

A 54 year-old female patient reported with swelling in right side of the face since one year. Patient claimed that mandibular right posterior teeth loosened and fell off, 1 year back. Personalhistory revealed that she chewed tobaccosince 5 years.

On examination there was a bony hard, tender swelling,on the right posterior mandible. The overlying skin was stretched,of normal colour and non-pulsatile. (Figure 1)

A single submandibular lymph node was palpable on right side which was 1cm in size, firm in consistency mobile and tender.

Vestibular obliteration, bicortical expansion from 44 to 48 region with egg shell crackling were noted. 46,47,48 was missing with grade 1 mobility in 45 which was tilted mesially.

Orthopantomograph showed a multilocular lesion in the right side of body ofmandible crossing the midline. It extended anterio-posteriorly from the right ramus upto 36, and superior-inferiorly from the crestto the lower border of mandible. The internal

structure was mixed radiopaque radiolucent showing varying patterns. It exhibited honeycomb and soap bubble appearance with respect to 36,37 region. 41,42 region showed wispytrabeculae intersecting at right angles giving a‘tennis racket’appearance, while 31,32 region shows thin trabeculae w resembling a ‘caricature of spider’ or ‘Spider Nevi appearance’.(Figure no. 2)

On the basis of radiographic findings in OPG, the provisional diagnosis was given as Odontogenic Myxoma,and the differential diagnosis was Ameloblastomaand Central Giant Cell Granuloma.

A CBCT with 3D reconstruction was done withFOV - 12 x 5 cm. CBCT findings revealed mixed hypodense and hyperdenselesion in right mandibular posterior region with ill-defined margins .Antero-posteriorly extending from 34 up to the ramus on the right side, lesion was crossing the midline, measuring approx.* 22.7 x 55.3mm.The internal structure was mixed multilocular with tennis racket appearance in 31, 32 region and honey comb appearance in 46,47 regions, spider nevi pattern (caricature of spider) in 41,42 region. The trabeculae were incomplete and wispy, expansion in bucco-lingual cortical plate was noted. Theinferior alveolar nerve canal was untraceable on right and left side. Thinning of the buccal and lingual cortical plate was seen on right side. The entire bony architecture seemed to be affected in the mandible, with change in the trabecular pattern on the left side. Root resorption was noted with 46.(Figure 3)

Provisional diagnosis according to CBCT findings was suggestive of Ameloblastoma ofmandible. Differential diagnosis was given as Odontogenic Myxoma, Central Giant cell granuloma. In Rarities,Browns Tumor of hypothyroidism and central hemangioma were included in differential diagnosis.

Afterincisional biopsy, histopathological examination revealed H&E-stained specimen of blood and its components, with no sign of malignant cells or other pathological cells. The punch biopsy of the lesion revealed characteristics consistent with cavernous hemangioma, without the presence of giant cells or malignancy.

Anenblock excision of the lesion including a safety margin of surrounding healthy bone and nutrient vessel ligation was done, along with reconstruction using bone graft and titanium plating.

Final specimen was sent for histopathological examination, the photomicrograph revealed the presence of bonytrabeculae and spicules between numerous vascular structures with muscular walls and dilated vascular spaces separated from one another by fibrous tissue.These features were suggestive of intraosseous cavernous haemangioma.

Postoperative OPG with 3 month follow up showed satisfactory healing without any possible relapse. (Figure 4)

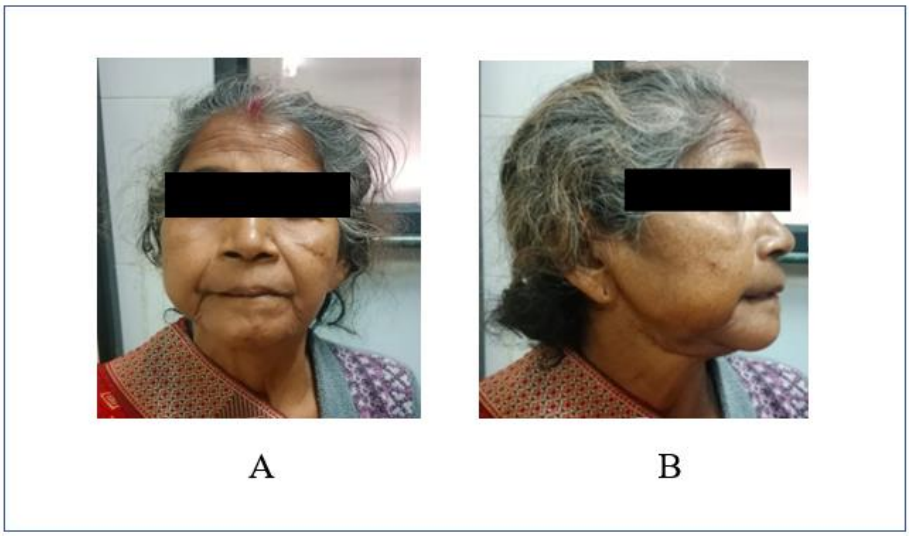

Figure 1 Facial profiles of patient. A Frontal view B Right lateral view

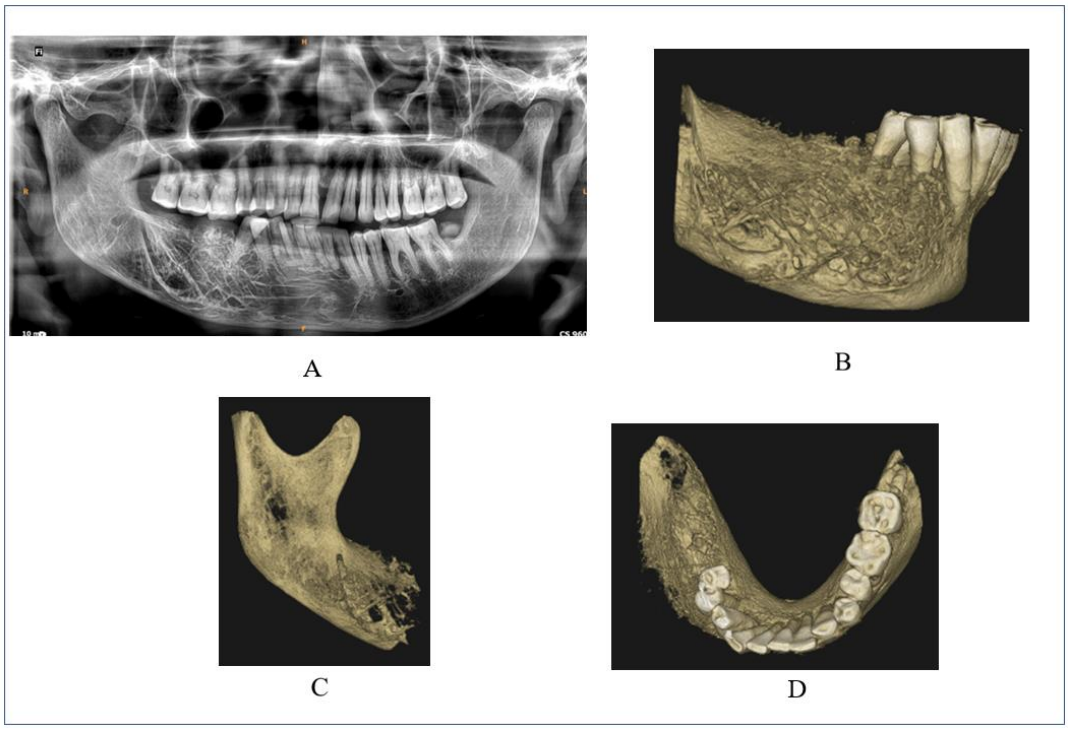

Figure 2: Preoperative radiographic evaluation. A Orthopantomogram; 3D rendered images of mandible: B buccal view of right posterior mandible. C buccal view of right ramus; D occlusal view

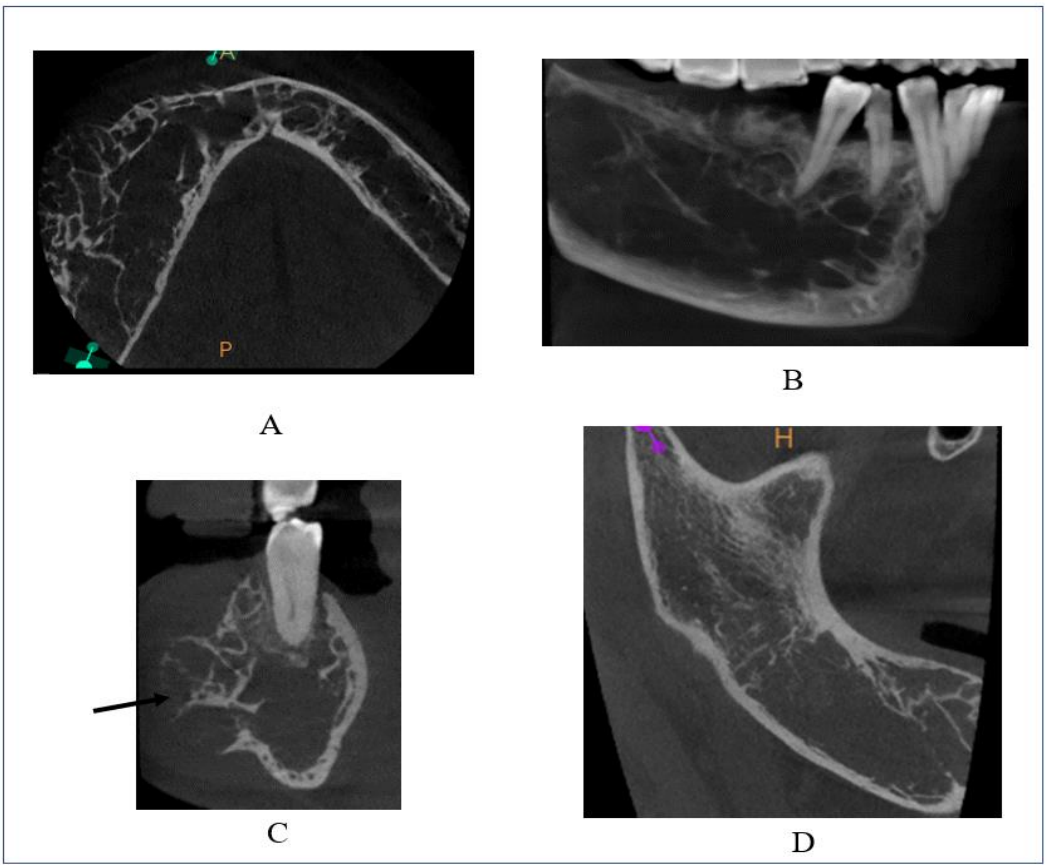

Figure 3: Cross sectional CBCT images of right side of mandible.
A: Axialimageof mandible (at level of mid-alveolar region) ;
B: Oblique sagittal image ;C: Coronal image in 45 region (arrow showing buccal expansion, thinning with interruption) ;
D : Oblique sagittal image of right ramus.

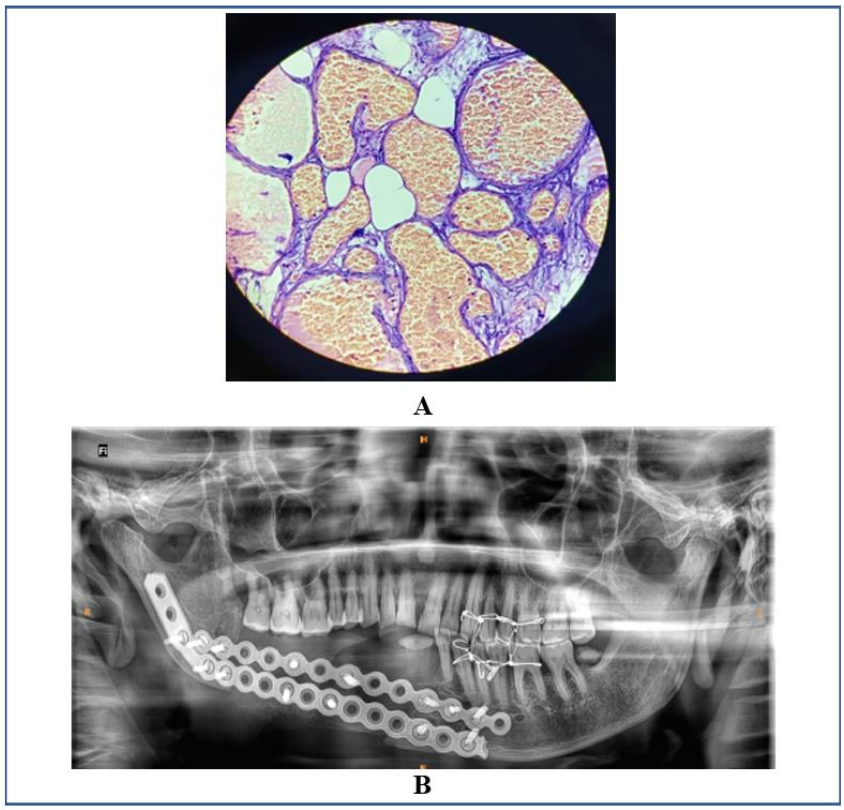

Figure 4 A:Histopathology (40x). B: Immediate post-operative orthopantomogram.

DISCUSSION

Hemangiomas are rare congenital vascular abnormalities seen in children and young adultsin second decade of life. They show higherfemale predilection, ratio being 2:1.[7]Cavernous hemangiomas primarily occur in the skull or spine, with occasional involvement of the maxilla and rarely in body of mandible. [8]

Two main theories have been postulated about its etiology one being it a true benign neoplasm, characterized by localized cell overgrowth forming a non-cancerous tumor, while the other suggests it to be hamartoma, arising from abnormal proliferation of mesodermal cells differentiating into endothelial cells. .[9,10]

Their elusive radiographic appearance among other multilocular lesions has consistently posed a diagnostic challenge for oral radiologists.

Langland et al [4]has described the presence of radiopaque striae with tube like arrangement associated with cavernous hemangioma, which was noted in the present case. Lesion reveals an area of altered radiodensity usually osteolytic with occasionally central radio-opaque areas and altered trabecular pattern. According to Worth trabecular pattern is similar to spoke wheel appearance radiating from center to periphery, which was also noted in the present case. Soap bubble or honeycomb-like appearance with multilocularosteolytic lesion similar to ameloblastoma can also be seen in central hemangioma which was again observed in the present case.[11, 12]Nagpal et al. in his case report described the variable appearance of lesion in a different projection.[12]Thus the CBCT findings manifested a whole plethora of radiographic features seen in the central hemangioma which was the most striking feature of the present case.This was a rarest of rare case where we could observe all the radiographic features described in the literature.

These capricious radiographic features of cavernous hemangioma,includes all the multilocular lesions as their differential diagnosis -ameloblastoma,OKC,giant cell lesion, myxoma, fibrous dysplasia, osteosarcoma, aneurismal bone cyst.[13]

Accurate diagnosis is the key in managing such vascular lesions so as to prevent unwanted complications. Management strategies depend on the size of lesions,location,patient's age and functional characteristics.Different management techniques include steroid injections, radiation therapy, embolization, curettage and surgical resection. Higher risk of bleeding in these lesions due to significant collateral circulation is the major complication in surgical resection of these lesions. In the present case enblock resection, with reconstructive surgery was done.[14]

CONCLUSION

Due to the complex clinical and radiological presentation definitive diagnosis can be reached only by careful elimination of other multilocular bony lesions. Therefore, effective management of cavernous hemangiomas requires a thorough multidisciplinary approach that considers the patient's

clinical presentation, imaging results, and the potential risks and benefits associated with the selected treatment method.

REFERENCES

1. Karjodkar FR. Textbook of Dental and Maxillofacial Radiology by Karjodkar. Jaypee Brothers Publishers; 2006.
2. Langland OE, Langlais RP, Preece JW. Principles of Dental Imaging. Lippincott Williams & Wilkins; 2002.
3. Wood NK, Goaz PW. Differential Diagnosis of Oral and Maxillofacial Lesions. 1997.
4. Langlais RP, Langland OE, Nortjé CJ. Diagnostic Imaging of the Jaws. 1995,
5. Yao K, Tang F, Min L, Zhou Y, Tu C. Multifocal intraosseoushemangioma. Medicine. 2019;98(2).,
6. Mattassi R, Loose DA, Vaghi M. Classification of Vascular Malformations. Hemangiomas and Vascular Malformations. 2009;:111-4.
7. Soto F, Alshehri S, Dekeister C, Lopez R. Active surveillance for intraosseous mandibular haemangioma. Case report and literature review. MédecineBuccaleChirurgieBuccale. 2017;23(2):90-4.
8. Dereci O, Acikalin MF, Ay S. Unusual intraosseous capillary hemangioma of the mandible. European Journal of Dentistry. 2015;9(3):438-41.,
9. Darrow DH, Greene AK, Mancini AJ, Nopper AJ. Diagnosis and Management of Infantile Hemangioma: Executive Summary. Pediatrics. 2015;136(4):786-91.,
10. Williams HJ, Wake MJ, John PR. Intraosseous haemangioma of the mandible: a case report. Pediatric Radiology. 2002;32(8):605.
11. Zlotogorski A, Buchner A, Kaffe I, Schwartz-Arad D. Radiological features of central haemangioma of the jaws. Dentomaxillofacial Radiology. 2005;34(5):292-6.
12. Nagpal A, Suhas S, Ahsan A, Pai K, Rao N. Central haemangioma: variance in radiographic appearance. Dentomaxillofacial Radiology. 2005;34(2):120-5
13. Dhiman NK, Jaiswara C, Kumar N, PatneShashikant C. U, Pandey A, Verma V. Central cavernous hemangioma of mandible: Case report and review of literature. National Journal of Maxillofacial Surgery. Vol 6; Issue 2 : Jul-Dec 2015.
14. Kim JE, Yi WJ, HeoMS, Lee SS, Choi SC, Huh KH. An unusual case of intraosseous vascular malformation of the maxilla mimicking fibrous dysplasia: a case report and literature review on imaging features of intraosseous vascular anomalies of the jaw. Dentomaxillofacial Radiology. 2014;43(7):20130400.