Case Report

Central cavernous hemangioma of mandible: Case report and review of literature

Department of Oral and Maxillofacial Surgery, Faculty of Dental Sciences, [1]Department of Pathology, Faculty of Modern Medicine, Institute of Medical Sciences, Banaras Hindu University, Varanasi, Uttar Pradesh, India

Neeraj Kumar Dhiman, Chandresh Jaiswara, Naresh Kumar, Shashikant C. U. Patne[1], Arun Pandey, Vishal Verma

ABSTRACT

Intraosseous hemangiomas are one of the rarest lesion of jaw bones (0.5–1%) occurring most commonly in vertebral column, skull bone, and rarely in mandible. Mainly occurs in the second decade of life with female: male predilection (2:1). Origin of hemangiomas is still debatable. World Health Organization considers it as a true benign neoplasm of vascular origin, and many authors believe it to be a hamartoma. It is very difficult to diagnose due to variable clinical and radiological features. A biopsy is not done on a routine basis due to a higher risk of hemorrhage. Management is very difficult because of massive vascular network in that region. Here, we are presenting a case report of a 14-year-old boy with intraosseous hemangioma of right body of mandible, which was treated with *en bloc* surgical resection of mandible and followed by reconstruction.

Address for correspondence:
Dr. Neeraj Kumar Dhiman, Department of Oral and Maxillofacial Surgery, Faculty of Dental Sciences, Institute of Medical Sciences, Banaras Hindu University, Varanasi, Uttar Pradesh, India.
E-mail: drnkdhiman@yahoo.co.in

Key words: Angiography, cavernous hemangioma, *en bloc* resection, mandible

INTRODUCTION

Hemangioma is a true benign neoplasm of endothelial origin. It is usually found in soft tissue and naturally follows rapid postnatal growth followed by slow spontaneous regression.[1,2] Intraosseous variant or cavernous hemangiomas are relatively rare and comprises 0.5–1% of all intraosseous tumors.[3] These are relatively rare in jaw bones as compared to vertebral column and skull bones. Cavernous hemangioma is mostly of congenital origin occurring frequently in mandible compared to maxilla and nasal bones.[4] In mandible, the body region is mostly affected, whereas some condylar tumors have also been reported.[5] 65% are found in the molar and premolar region. Sex predilection is 2:1 (female: male) with a peak incidence between the second and fifth decade of life.[1,6]

Access this article online

Quick Response Code:

Website: www.njms.in

DOI: 10.4103/0975-5950.183866

Origin of cavernous hemangioma is debatable. According to Shira and Guernsey, it is a true benign neoplasm as a result of endothelial proliferation which differentiates into blood vessels. Some authors suggest it to be a hamartomatous lesion which arises from the proliferation of mesoderm that undergoes endothelial differentiation which is further localized and vascularized.[7,8]

Mostly asymptomatic but may present with symptoms such as discomfort, pulsation, bluish slow growing mass, compression of surrounding structures, mobile teeth, and hemorrhage.[6] Orthopantomogram (OPG), computed tomography (CT), and magnetic resonance imaging (MRI) are mostly done for a diagnostic purpose. The correct diagnosis of cavernous hemangioma is

How to cite this article: Dhiman NK, Jaiswara C, Kumar N, Patne SC, Pandey A, Verma V. Central cavernous hemangioma of mandible: Case report and review of literature. Natl J Maxillofac Surg 2015;6:209-13.

relatively challenging due to similar radiologic features of ameloblastoma, odontogenic myxoma, fibrous dysplasia, and aneurysmal bone cyst.[9] Here, comes the role of history, clinical features, radiographs, and angiography in making a final diagnosis of hemangioma.

In this article, we report a rare presentation of cavernous hemangioma of right body of mandible, diagnosed and managed surgically at our unit.

Case Report

A 14-year-old boy reported to our unit of oral and maxillofacial surgery with the chief complaint of swelling on the right side of the lower face past 1 year. History of present illness revealed the sudden appearance of a swelling in the right side of the face which gradually progressed in shape and size. Swelling was associated with nonradiating dull pain. After 5–6 months, swelling size became stagnant with a lower intensity of pain. At present, he experiences a hard swelling which is painful on the application of pressure. No relevant medical history and history of spontaneous bleeding or paresthesia was given.

Extraoral examination revealed asymmetry on the right side of the face. Swelling was approximately 4 cm × 3 cm in size and present over the right mandibular body with diffuse margins extending from the right angle of mouth to the angle of mandible. On palpation, a bony hard, smooth, tender, and diffuse swelling was present in the lateral aspect of the right mandibular body involving the inferior border.

Intraorally, mouth opening was normal with satisfactory occlusion, mandibular right third molar was embedded, the second molar was lingually tilted, central incisor was with Grade 1 mobility, and canine was present labial to the incisors [Figure 1]. There was diffuse hard swelling obliterating the buccal vestibule, extending from mandibular right first premolar to retromolar region. Only buccal cortical expansion was present with mild blanching of mucosa without any pulsations.

Radiographic analysis was done on the basis of OPG and CT face. OPG exhibited increase in the vertical dimension of the right side of mandibular body, coarse trabeculation, haziness, and increased radio-opaque striations from center to periphery extending from the first premolar to ramus region. There was flaring of roots of premolars and molars of the right side and root resorption of central and lateral incisors on either side of mandible [Figure 2].

CT scan revealed an ill-defined, heterogeneous multiseptated osteolytic lesion of size (6.2 cm × 2.7 cm × 4.8 cm) involving body and ramus of right mandible with honeycombed appearance and having cortical bone insufflations along with characteristic sun burst appearance of periosteum as evident by spindles radiating toward periphery. There is a mild expansion of diploic space of right mandible with thinning and multiple cortical breaks of outer cortical plate [Figure 3a and b]. CT angiography of face did not reveal any feeding vessels to the lesion.

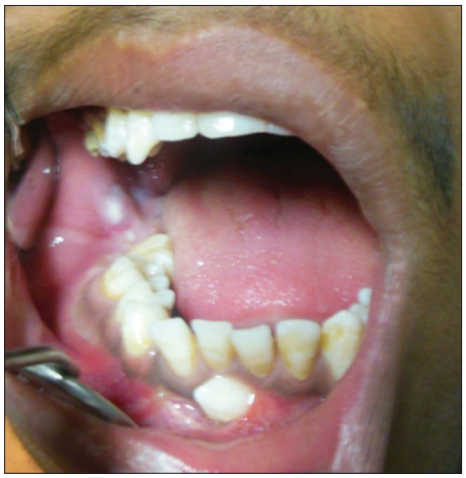

Figure 1: Intraoral view of the lesion

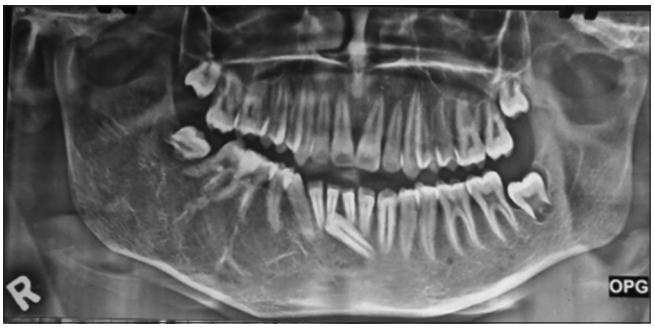

Figure 2: Panoramic view of the lesion showing extension and involvement of right side of mandible

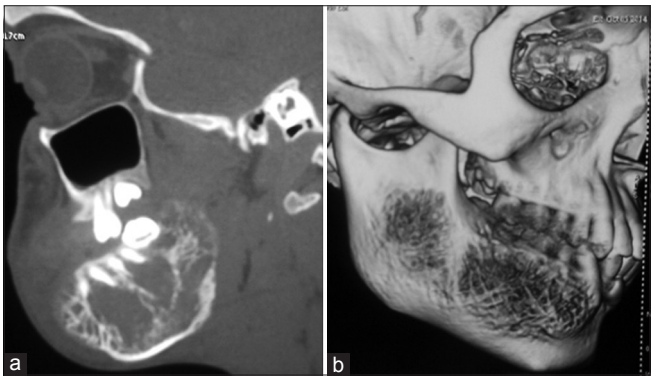

Figure 3: (a) Sagittal view showing the involvement of right mandibular body and ramus. (b) Three-dimension computed tomography showing multiple septa and cortical breaks in the outer cortex of body and ramus region

After fine needle aspiration cytology, which yields little hematic material and no apparent cellularity, a biopsy was planned and histopathology report came as cavernous hemangioma of right body of the mandible [Figure 4]. Surgery was planned, under general anesthesia. First, curettage of the lesion was attempted but because of perforations present on the lingual cortex of ramus, segmental resection of right body of mandible was performed combined with additional L-shaped osteotomy over ramus and premolar region to achieve safe margins followed by immediate reconstruction with 2.5 mm reconstruction titanium locking plate (used as a spacer for further grafting) [Figure 5a and b]. Blood transfusion was done postoperatively. Specimen was sent for microscopic examination which remains same as cavernous hemangioma of mandible. The patient is under regular follow-up and being planned for bone grafting.

Discussion

Clinical features

Mostly asymptomatic and may take months for the symptoms to become obvious when the patient seeks treatment. Lesion occurs as firm, painless bony swelling which may be minimal or causing gross facial asymmetry and sometimes associated with pulsation.[7,10,11] Pain and paresthesia are not characteristic features but may be associated with the swelling. Intraorally, common findings are obliteration of vestibule, expansion of buccal or lingual cortex, mobility, and displacement of teeth. Occasionally, root resorption also occurs. Supra eruption, premature exfoliation of primary teeth, and early eruption of permanent teeth also have been reported.[12,13]

Radiographic features

Radiographic features of cavernous hemangioma simulate many other lesions of the jaw, so definitive diagnosis based on it is difficult. The presence of parallel or tube-like arrangement of radio-opaque striae is an important feature of cavernous hemangioma as described by Langland *et al.* lesion reveals an area of altered radiodensity usually osteolytic with occasionally central radio-opaque areas and altered trabecular pattern.[14] According to Worth trabecular pattern is similar to spokes of wheel, radiating from center to periphery. Multicystic osteolytic areas give a soap bubble or honeycomb-like appearance.[15] Nagpal *et al*. in his case report described the variable appearance of lesion in a different projection.[16]

Periphery shows well-defined or ill-defined corticated area with scalloped margin. Due to the presence of a variable degree of radiolucency cavernous hemangioma may be multilocular or unilocular and may give sunburst and tennis racket appearance also.[17]

Due to this highly variable radiographic picture differential diagnosis of cavernous hemangioma may include - (1) ameloblastoma, (2) giant cell lesion, (3) myxoma, (4) dentigerous cyst, (5) fibrous dysplasia, (6) osteosarcoma, (7) aneurismal bone cyst, and (8) granuloma. Occasionally, indistinguishable from vascular anomalies such as aneurysms and shunts.[14,17]

CT angiography reveals feeder vessels and vascular abnormality thus plays an important role in reaching to a definitive diagnosis.[18]

Histopathological examination

On the basis of histology, hemangioma has been classified into (1) capillary, (2) cavernous, and (3) mixed variant.[19]

Endothelial cells proliferate and form a plexiform pattern of vascular space. The thin-walled cavernous spaces are lined by a single layer of endothelial cells interspersed among bony trabeculae. Hitzrot[20] describes the development of hemangioma in three stages (1) early: Highly vascular, (2) intermediate: Exhibits blood clotting, and (3) terminal: Various stages of ossification.

Treatment modalities

Various treatment modalities for cavernous hemangioma have been described in literature based upon,

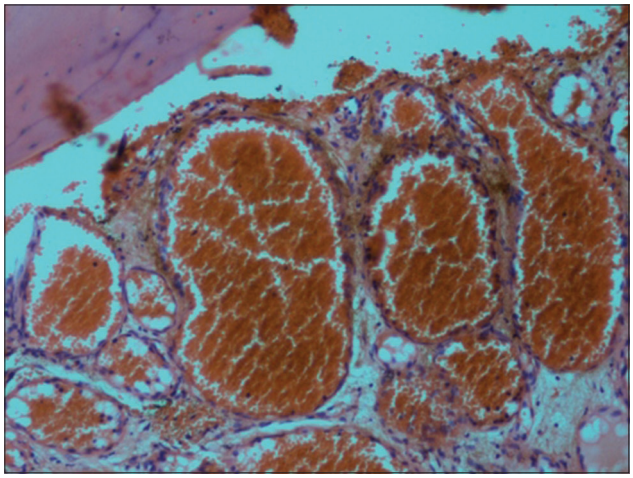

Figure 4: Microscopic picture of the lesion

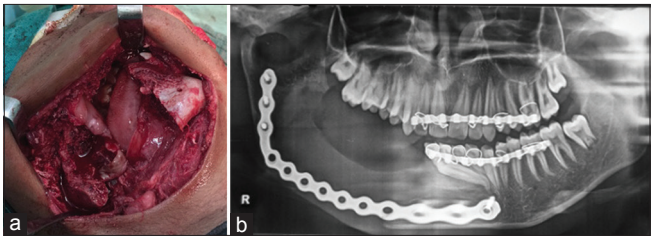

Figure 5: (a) Intraoperative view defect after resection. (b) Postoperative view showing bridging of the defect by reconstruction plate

(1) hemorrhage control, (2) complete eradication of the lesion, and (3) prevent recurrence.[10,13]

These include: (1) noninvasive radiotherapy, (2) intralesional injection of sclerosing agents and embolization, (3) curettage and radiation, and (4) resection followed by osseous reconstruction.

Treatment should be based upon clinical findings, patient's age, and medical history. Radiation is given for inaccessible lesions. According to Jaffe, growth of lesion may be controlled by radiotherapy, but osseous deformity can only be corrected by surgery.[21] Macnash and Owen, have reported cases which treated successfully by radiation alone (500R–3300R).[22] Wilde *et al*. described cavernous hemangioma as radioresistant, and associated complications with radiotherapy are damage to condylar growth, developing teeth and salivary glands.[23]

Intralesional injection of sclerosing agents such as boiling water, sodium morrhuate, and sodium tetradecyl sulfate have been tried for extensive lesions. These acts as tissue irritant and thrombogenic agents but have limited application in the case of osseous lesions.[12] Embolization of major afferent vessels feeding cavernous hemangioma is also treatment when the patient is not an ideal candidate for surgery.[24]

Surgery alone or in combination with embolization still remains the best treatment option for cavernous hemangioma.[8,13] Conservative surgical methods include aspiration of intraosseous lesion. The size of the lesion reduces due to a reduction in vascularity, fibrosis, and reossification.[7] Surgery includes either curettage or radical excision of a segment of jaw followed by immediate bone graft reconstruction. In curettage, buccal plate is osteotomized. and hemorrhagic tissue is removed while preserving the continuity of the jaw. Block resection followed by immediate iliac crest reconstruction is the most effective and safest treatment modality as suggested by Ladow and Mcfall.[25]

As blood loss during surgery is anticipated, adequate replacement for blood transfusion should be planned prior to the surgery.[25] Patient's attendant should be informed, and patient's blood group determination and cross matching should be done. Hemoglobin level assessment before and after surgery is necessary.

In this case, radiotherapy was avoided due to the young age of the patient and associated complications of radiation. Sclerosing agent did not have much role due to intraosseous nature of the lesion. Thus, segmental resection followed by reconstruction with 2.5 mm reconstruction was planned. The result was satisfactory; the patient is under regular follow-up and being planned for bone grafting.

Conclusion

Thus, due to the complex presentation of cavernous hemangioma, step by step approach toward a definite diagnosis should be made by excluding other bony lesions of same characteristics. Microscopic examination along with CT angiography is the gold standard for making a definitive diagnosis. Treatment modality should be carefully planned based upon patient's age, clinical features, extent of the lesion, and systemic medical status.

Financial support and sponsorship
Nil.

Conflicts of interest
There are no conflicts of interest.

References

1. Alves S, Junqueira JL, de Oliveira EM, Pieri SS, de Magalhães MH, Dos Santos Pinto D Jr, *et al*. Condylar hemangioma: Report of a case and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2006;102:e23-7.
2. Williams HJ, Wake MJ, John PR. Intraosseous haemangioma of the mandible: A case report. Pediatr Radiol 2002;32:605-8.
3. Beziat JL, Marcelino JP, Bascoulergue Y, Vitrey D. Central vascular malformation of the mandible: A case report. J Oral Maxillofac Surg 1997;55:415-9.
4. Moore SL, Chun JK, Mitre SA, Som PM. Intraosseous hemangioma of the zygoma: CT and MR findings. AJNR Am J Neuroradiol 2001;22:1383-5.
5. Whear NM. Condylar haemangioma – A case report and review of the literature. Br J Oral Maxillofac Surg 1991;29:44-7.
6. Cheng NC, Lai DM, Hsie MH, Liao SL, Chen YB. Intraosseous hemangiomas of the facial bone. Plast Reconstr Surg 2006;117:2366-72.
7. Shira RB, Guernsey LH. Central cavernous hemangioma of the mandible: Report of case. J Oral Surg 1965;23:636-42.
8. Sadowsky D, Rosenberg RD, Kaufman J, Levine BC, Friedman JM. Central hemangioma of the mandible. Literature review, case report, and discussion. Oral Surg Oral Med Oral Pathol 1981;52:471-7.
9. Fernández LR, Luberti RF, Domínguez FV. Radiographic features of osseous hemangioma in the maxillo-facial region. Bibliographic review and case report. Med Oral 2003;8:166-77.
10. Bunel K, Sindet-Pedersen S. Central hemangioma of the mandible. Oral Surg Oral Med Oral Pathol 1993;75:565-70.
11. Mohnac AM, Smith JR. Central hemangioma of the mandible: Report of case. J Oral Surg 1967;25:455-9.
12. Hayward JR. Central cavernous hemangioma of the mandible: Report of four cases. J Oral Surg 1981;39:526-32.
13. Yih WY, Ma GS, Merrill RG, Sperry DW. Central hemangioma of the jaws. J Oral Maxillofac Surg 1989;47:1154-60.
14. Langland OE, Langlais RP, McDavid WD, Delbalso AM, editors. Multilocular radiolucencies. In: Panoramic Radiology. 2nd ed. Philadelphia, PA: Lea and Febiger; 1989. p. 288-90.
15. Worth HM, editor. Benign tumors of jaw. In: Principles and Practice

of Oral Radiologic Interpretation. Chicago, Ill: Yearbook Medical Publishers, Inc.; 1969. p. 522-8.
16. Nagpal A, Suhas S, Ahsan A, Pai K, Rao N. Central haemangioma: Variance in radiographic appearance. Dentomaxillofac Radiol 2005;34:120-5.
17. Zlotogorski A, Buchner A, Kaffe I, Schwartz-Arad D. Radiological features of central haemangioma of the jaws. Dentomaxillofac Radiol 2005;34:292-6.
18. Guibert-Tranier F, Piton J, Riche MC, Merland JJ, Caille JM. Vascular malformations of the mandible (intraosseous haemangiomas). The importance of preoperative embolization. A study of 9 cases. Eur J Radiol 1982;2:257-72.
19. Thoma KH, Goldman HM. Bone tumors of the jaws. In: Gorlin RJ, Goldman HM, editors. Thoma's Oral Pathology. 6th ed. St. Louis, Mo: CV Mosby Company; 1971. p. 564-6.
20. Hitzrot JM. Haemangioma cavernosum of bone. Ann Surg 1917;65:476-82.
21. Jaffe HL. Tumors and Tumorous Conditions of the Bones and Joints. Philadelphia, PA: Lea and Febiger Publishers; 1953. p. 236-7.
22. Macnash JD, Owen M. Central cavernous hemangioma of mandible: Report of a case. J Oral Surg 1972;30:293-5.
23. Wilde NJ, Tur JJ, Call DE. Hemangioma of the mandible: Report of case. J Oral Surg 1966;24:549-52.
24. Kaneko R, Tohnai I, Ueda M, Negoro M, Yoshida J, Yamada Y. Curative treatment of central hemangioma in the mandible by direct puncture and embolisation with n-butyl-cyanoacrylate (NBCA). Oral Oncol 2001;37:605-8.
25. Ladow CS, Mcfall TA. Central hemangioma of the maxilla, with von hippel's disease: Report of a case. J Oral Surg Anesth Hosp Dent Serv 1964;22:252-9.