

# Intraosseous Hemangioma: A Case Report and Review of Literature

Yogendra Singh Chauhan, Srijon Mukherji

## ABSTRACT

Intraosseous vascular lesions are rare conditions, comprising only 0.5 to 1% of all intraosseous hemangioma tumors. They mainly occur in the second decade of life especially in female. The most common locations are the vertebral column and skull; nevertheless, the mandible is quite rare location. Hemangiomas are benign vasoformative neoplasms of endothelial origin. However, the origin of central hemangioma is debatable. Cavernous hemangioma produces dilemma in diagnosis with central giant cell tumor, aneurysmal bone cyst, ameloblastoma, cystic lesion such as residual cyst, keratocyst and fibro-osseous lesions, such as fibrous dysplasia being frontier in clinical diagnosis. Here, we report a 6 years male with cavernous hemangioma of mandible.

**Keywords:** Cavernous hemangioma, Mandible, Male.

**How to cite this article:** Chauhan YS, Mukherji S. Intraosseous Hemangioma: A Case Report and Review of Literature. Int J Head Neck Surg 2013;4(2):66-69.

**Source of support:** Nil

**Conflict of interest:** None declared

## INTRODUCTION

Hemangioma is benign vasoformative neoplasm of endothelial origin.<sup>1,2</sup> Its natural course includes a rapid post-natal growth followed by a slow spontaneous regression which may take several years.<sup>2</sup> It is usually located in soft tissue. Intraosseous hemangioma is quite rare, comprising less than 1% of all intraosseous tumors.<sup>1,3</sup> It mainly occurs in the vertebral column. Mandible is very infrequent location with male to female ratio being 1:2 and the peak incidence is between the second and fifth decade of life.<sup>3</sup> Its origin is still debatable with some believe it is a true neoplasm, whereas others state it is a hamartoma resulting from proliferation of intraosseous mesodermal cells that undergo endothelial differentiation. It is usually asymptomatic although may present signs and symptoms including a slow growing bluish mass, discomfort, pulsatile sensation and mobile teeth.<sup>3</sup>

Panoramic radiographs computed tomographic (CT) scan and Magnetic resonance imaging (MRI) are the most useful radiographic studies. Radiological findings include:<sup>4</sup>

1. Unilocular rounded lesion, of varying size and appearance, resembling a cyst.
2. Well-defined cavity with sclerotic rims and anarchic inner trabeculation.
3. Bone trabeculae radiating from the center to the periphery of the lesion.

4. Honeycombed or sunburst appearance with spindles radiating toward the periphery, differentiating hemangioma from ameloblastoma.
5. A radiodense area is possible but represents a rare condition.

CT scan allows clear visualization of cortical involvement while MRI shows blood flow, if present, as well as the relationship with surrounding soft tissue.<sup>4</sup> Differential diagnosis include:<sup>4</sup>

- Odontogenic tumors
  - *Ameloblastoma*: Defined by histological ameloblastic differentiation and absence of sunrise radiological image.
  - *Myxoma*: Presents spindle cells scattered into mucoid stroma filled with mucopolysaccharides.
- Cystic lesions
  - *Odontogenic cyst*: Lacks radiological trabeculation
  - *Aneurysmal bone cyst*: Characterized by its fast growth and its bad defined edges filled with liquid in the radiological exploration.
- Fibrous lesion
  - *Fibrous dysplasia*: Painful lesion that present reactive bone formation in a fibrous origin in contrast to moderately remodelated laminar bone of the hemangioma.

## CASE REPORT

In this article, we report the case of a patient diagnosed with hemangioma histopathologically with unusual age, sex, radiographic features and location creating huge diagnostic dilemma toward management of particular case.

A 6-year-male child presented to our institute with complaints of right facial swelling from 3 months. The patient was apparently healthy and his past medical history was non-contributory. Extraoral examination revealed a single diffuse swelling on right side of cheek. The color of overlying skin was normal (Figs 1 and 2).

Intraoral examination showed a bluish colored swelling located in the right mandibular body and ramus with variegated consistency. There were no other associated symptoms.

A orthopantomogram revealed a 7 × 7 cm ill-defined radiolucent image involving right mandibular ramus, coronoid process, and body affecting upto right lateral incisor

(Fig. 3). Differential diagnosis included radicular cyst, solitary bone cyst, ameloblastoma, myxoma, central giant cell tumor, aneurysmal bone cyst and bone hemangioma. Given these findings a 3D-CT scan was performed revealing  $7 \times 5 \times 2.5$  cm well-defined lesion with bone trabeculation, lytic voids of cortex and periostotic reaction with cortices expansion of mandible. There was remarkable alteration of dental roots and inferior alveolar canal (Fig. 4).

A biopsy performed elsewhere revealed central giant cell tumor aneurysmal bone cyst.

Patient was prepared to surgical resection of the lesion with reconstruction as curettage in our provisional diagnosis have high incidence of recurrence reaching up to 70% which being unimpressive to surgeon and patient putting huge economical strain on family and society.

Hemimandibulectomy with whole free rib graft reconstruction was planned by lip-split mandibulotomy approach under general anesthesia. After giving incision, buccal flap was raised to exposed whole lesion with careful dissection to free lingual aspect from skull base leaving articular cartilage. Whole rib graft including costochondral junction was harvested with preservation of cuff of periosteum

and perichondrium at the costochondral junction for preserving growth center. During surgery, specimen had high bleeding tendency which prompted us our diagnosis in favor of intraosseous hemangioma (Figs 5 to 9).

The surgical specimen was described macroscopically as right mandible measuring  $7 \times 5 \times 2.5$  cm with gross resorption and thinning of buccal and lingual cortical plates, body of mandible is spongy and filled with blood clot. Microscopic examination reveals multiple, large, dilated, sinusoidal spaces, lined by flattened endothelial cells. The spaces filled with RBCs. There are profuse extravasated RBCs, multiple bony trabeculae, osteoids and muscle tissue. At places thin cavernous space ramify with one another. No giant cell are present in the given specimen which highlights to dismiss first biopsy performed elsewhere with surgical clearance around 0.5 to 1 cm. Histopathological diagnosis of intraosseous hemangioma was made.

## DISCUSSION

The most frequent location is the molar—premolar region.<sup>5</sup> Pathogenesis is still debatable and several theories are postulated. Some authors describe hemangioma as congenital



Fig. 1: Preoperative extraoral view

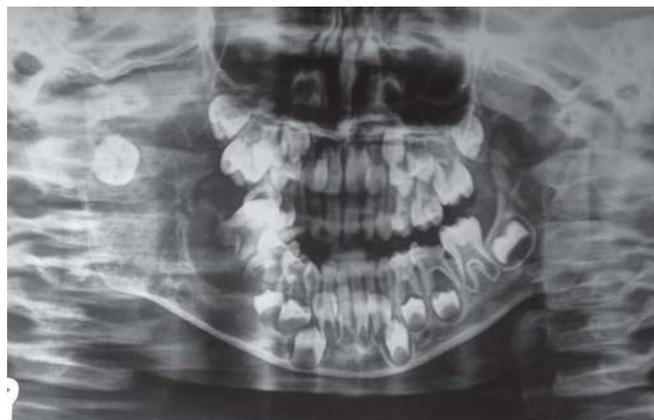


Fig. 3: Orthopantomogram



Fig. 2: Preoperative oral view

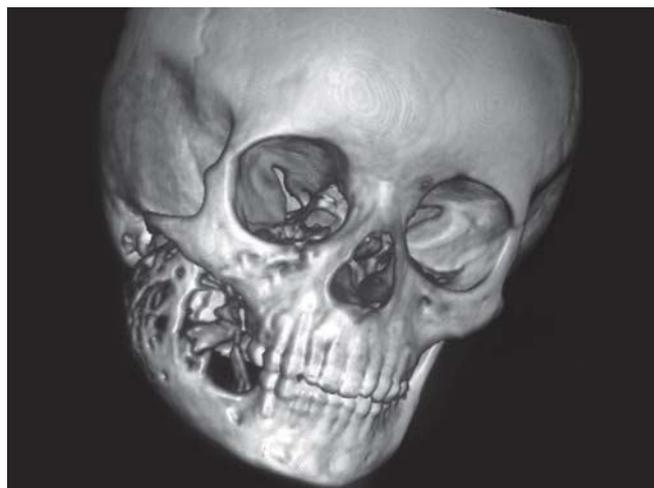


Fig. 4: 3D CT scan

lesions whereas others believe that inferior dental canal is origin of the lesion, based on its widening in the majority of these patients.<sup>3,5</sup>

The initial diagnosis is usually complicated because of the absence of symptoms and nonspecific radiological findings. The CT scan allows clear visualization of cortical involvement and to define extension of hemangioma and its relationship with surrounding soft tissue.<sup>6</sup> The classical feature is the 'polka-dot' appearance with cortical expansion.<sup>7</sup>

Preoperative arteriography is usually unnecessary because a vascular flow cannot be identified in majority of cases. Nevertheless it could be performed, if confirmed with preoperative diagnosis of hemangioma for presurgical embolization in big lesions to minimize surgical bleeding.<sup>8</sup>

Biopsy is formally contraindicated because of the high risk of bleeding.

*There are two types of hemangioma:* Peripheral and central. Peripheral hemangioma originated in periostic vessels that grow into medullar bone, while central hemangioma is originated into the medullar bone and grow toward the cortical bone.

*Treatment is indicated in conditions:* Esthetic disfigurement, repetitive bleeding and palpable mass.

*Therapeutic alternatives include:* Surgery, radiotherapy, curettage and embolization.

Radiotherapy is useful to reduce tumoral volume. Nevertheless, it has a lot of adverse effects, such as damage to normal adjacent tissue growth, residual scarring and malignization. So, radiotherapy is considered an unacceptable therapeutic option.

Simple curettage may lead to an uncontrollable bleeding as well as an incomplete excision of the lesion leading to high chance of future recurrence.

Percutaneous embolization has been defended by several authors, although technical risks are greater than benefits.<sup>9</sup>

Systemic or intralesional corticosteroids can also be used along with angiogenesis inhibitors, such as interferon in selected cases.<sup>2</sup>

Surgical excision with reconstruction of mandible remains the preferred treatment. Prognosis after complete excision is excellent and recurrence is usually rare.<sup>10,11</sup> In our case,

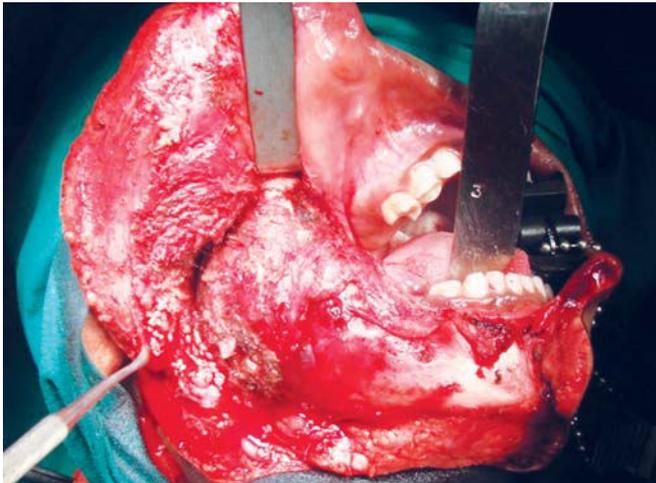


Fig. 5: Exposed view

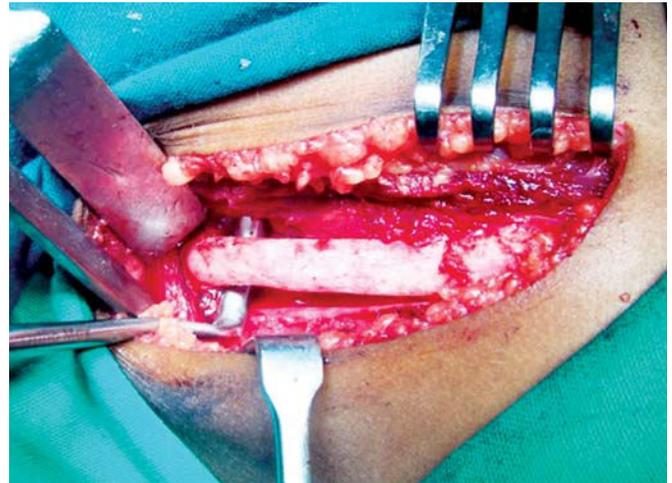


Fig. 7: Rib harvest

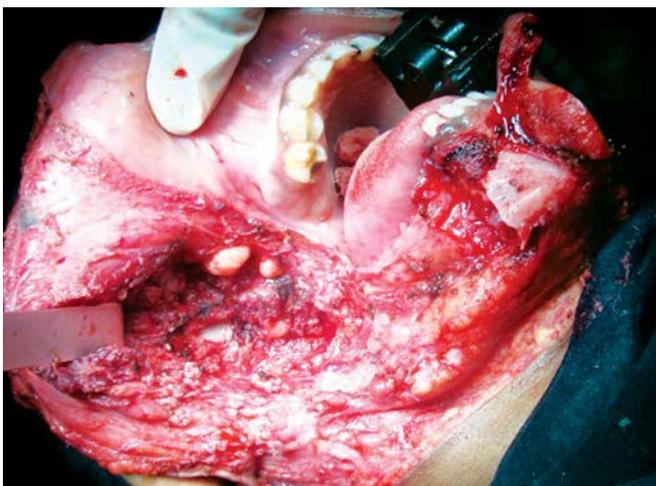


Fig. 6: Removal of tumor

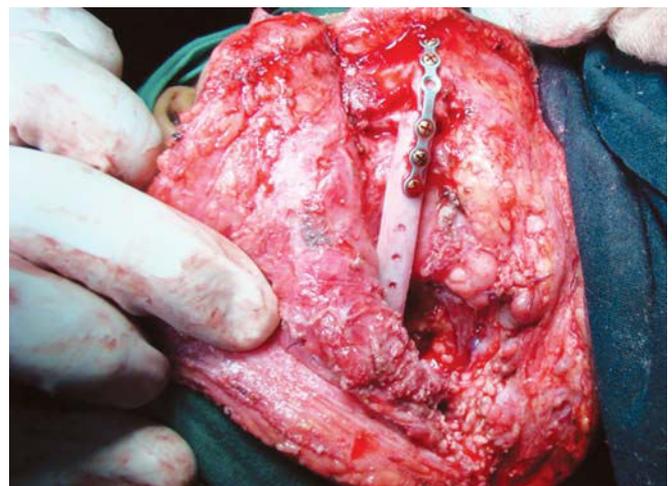


Fig. 8: Reconstruction



**Fig. 9:** Immediate postoperative view

hemimandibulectomy with sufficient surgical margin and reconstruction was done using whole free rib graft.

## REFERENCES

1. Alves S, Junquiera JL, De Oliveira EM, Pieri SS, De Magalhaes 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 Nov;102(5):e23-27.
2. Williams HJ, Wake MJ, John PR. Intraosseous haemangioma of the mandible: A case report. *Pediatr Radiol* 2002 Aug;32(8):605-08.
3. Cheng NC, Lai DM, Hsie MH, Liao SL, Chen YB. Intraosseous hemangioma of the facial bone. *Plast Reconstr Surg* 2006 Jun;117(7):2366-72.
4. Fernandez LR, Luberti RF, Dominguez FV. Radiographic features of osseous hemangioma in maxillofacial region. Bibliographic review and case report. *Med Oral* 2003 May-Jul;8(3):166-77.
5. Drage NA, Whaites EJ, Hussain K. Hemangioma of the body of the mandible: A case report. *Br J Oral Maxillofac Surg* 2003 Apr;41(2):379-81.
6. Sweet C, Silbergleit R, Mehta B. Primary intraosseous hemangioma of the orbit: CT and MR appearance. *Am J Neuroradiol* 1997 Feb;18(2):379-81.
7. Zlotogorski A, Buchner A, Kaffe I, Shwartz-Arad D. Radiological features of central hemangioma of the jaws. *Dentomaxillofac Radiol* 2005 Sep;34(5):292-96.
8. Kaneko R, Tonhai L, Ueda M, Negoro M, Yoshida J, Yamada Y. Curative treatment of central hemangioma in the mandible by direct puncture and embolisation with nbutyl-cyanoacrylate (NBCA). *Oral Oncol* 2001;37:605-08.
9. Liu D, Ma X, Assesment of efficacy of endovascular embolization for central arteriovenous malformation (AVM) in the jaw. *Zhonghua Kou Qiang Yi Xue Za Zhi* 2002;37:340.
10. Behnia H, Ghodoosi I, Motamedi MH, Khojasteh A, Masjedi A. Treatment of arteriovenous malformations: Assessment of 2 techniques-transmandibular curettage versus resection and immediate replantation. *J Oral Maxillofac Surg* 2008;66:2557-65.
11. Shum JW, Clayman L. Resection and immediate reconstruction of a pediatric vascular malformation in the mandible: Case report *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2010;109:517-24.

## ABOUT THE AUTHORS

### Yogendra Singh Chauhan (Corresponding Author)

Surgical Fellow, Department of ENT and Head and Neck Surgery Calcutta, Institute of Maxillofacial Surgery and Research, Kolkata West Bengal, India, e-mail: dr.yogendraredd@yahoo.com

### Srijon Mukherji

Consultant, Department of Maxillofacial, Calcutta Institute of Maxillofacial Surgery and Research, Kolkata, West Bengal, India