

# SURGICAL TREATMENT OF INTRAOSSEOUS HEMANGIOMA OF THE MANDIBLE: A CASE REPORT AND REVIEW OF LITERATURE

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## Abstract

Intraosseous vascular lesions are unusual conditions, comprising only 0.5% to 1% of all intraosseous tumors. These benign vascular malformations present variable and atypical radiographic appearances and because of this variation, no pathognomonic sign exists that can be used to make a definite clinical diagnosis of such pathological entities. In this paper, we discuss the case report of a 59 years old female who reported with a chief complaint of swelling in the anterior mandible since last 3-4 months. The purpose of this review article is to stress on the early diagnosis and appropriate surgical treatment which is crucial for accurate management of such lesions.

**Key words:** Intraosseous hemangioma, Multi-locular, Segmental Mandibulectomy, Fibular microvascular free flap.

## Introduction

Vascular anomalies in the Head & Neck region represent less than 1% of all bony tumors. They are classified into Hemangioma and Vascular malformation.<sup>1</sup> Hemangioma is the most common cutaneous tumor of infancy which is characterized by rapid proliferative growth followed by slow spontaneous involution. Unlike hemangioma, a vascular malformation are noted at the time of birth, enlarge proportionately with the growth of the child and does not undergo resolution spontaneously.<sup>1</sup> The true origin of Hemangioma is subject to disagreement in the literature.<sup>2</sup>

Intraosseous hemangioma was first described by Stanley in 1849, including central and peripheral intraosseous hemangiomas.<sup>3</sup> The peripheral type originates in the vessels of the periosteum with a path of growth towards the medullary bone. On the other hand, central hemangioma originates in the medullary bone extending towards the bony cortex.

Intraosseous hemangioma is an extremely rare condition, constituting 0.7% of all intraosseous tumors with rare occurrence in the jaws.<sup>3</sup> In India, less than 50 cases of intraosseous hemangioma of the mandible have been reported so far, which further emphasizes on the rarity of this lesion.<sup>4</sup> Intraosseous hemangioma occurs with the highest frequency in children while it recedes following the adolescent years. The female to male ratio is 2:1 and the peak incidence is between the second and fifth decade of life.<sup>5,6</sup> They are usually found in the vertebral column and skull bones, and the facial bones are rarely affected.<sup>7,8</sup>

These osseous lesions have been referred to as "the great mimicker" because they can produce many different radiographic metaphors.<sup>5</sup> As the radiographic presentation can be misleading, biopsy of such vascular lesions or even a simple tooth extraction without a prior knowledge can result in catastrophic hemorrhages leading to death.

Here, we report a case of central hemangioma occurring in the anterior mandible with its radiographic presentation of sunburst appearance and a brief review of the surgical treatment.

## Case Report

A 59 years old female patient reported to our unit with the chief complaint of swelling in the lower anterior vestibular region since past 3-4 months. The swelling was initially small in size and gradually grew to its present size. The patient was not suffering with any systemic illness nor had any known drug allergy. (figure 1)



Figure 1: - Preoperative profile view

## Clinical Examination

On clinical examination there was no significant finding extraorally. Intraoral examination revealed a solitary well defined dome shaped swelling in the labial mucosa of the lower lip region extending from distal aspect of 32 till the distal aspect of 43 region and from the labiokingival sulcus till the crest of the residual alveolar ridge continuing lingually till the linguokingival sulcus measuring approximately 4.0 x 3.0 cm in its greatest dimensions.

The surface of the lesion was smooth and shiny with mild bluish hue. On palpation, the swelling was non-tender, compressible but non-reducible and soft in consistency. (figure 2)

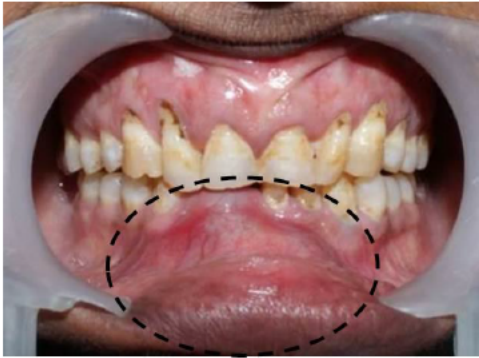


Figure 2: - Intraoral Preoperative view demonstrating the extent of the lesion in anterior region of the mandible

### Radiographic Examination

Panoramic radiograph demonstrated a honey-comb pattern of bone in the anterior body region of the mandible extending from tooth 46 till tooth 35. The periphery appeared smoothly demarcated from the adjacent unaffected bone but failed to reveal the expansile nature of the lesion. (figure 3)



Figure 3: - OPG reveals multiocular radiolucency in anterior mandible extending upto 35.

Mandibular occlusal radiograph revealed a sunburst appearance of the lesion in the anterior mandible with marked expansion and internal septations. (figure 4)



Figure 4: - Mandibular occlusal view demonstrate classical soap bubble appearance with multiple septae

CT Scan of the mandible revealed a non-expansile osteolytic lesion in the right body of the mandible causing erosion of the cortex of alveolar process with an intramedullary soft tissue component. An Ultrasound

guided biopsy of anterior mandible suggested a reactive/vascular benign lesion. Carotid angiogram failed to identify vessel associated with the lesion. (figure 5)



Figure 5: - CT scan suggested mandibular erosion extending from 43 to 35 region

Based on the clinical and laboratory investigations, a differential diagnosis of central giant cell granuloma, ameloblastoma, aneurismal bone cyst, odontogenic myxoma and benign vascular lesion.

Treatment plan consisted of enbloc resection of the lesion via segmental mandibulectomy and primary reconstruction using fibular microvascular free flap.

### Surgical Procedure

Under nasotracheal intubation, general anesthesia was achieved. Following skin preparation with an antiseptic, the intended surgical field was draped.

Visor incision was made through the skin and subcutaneous tissue in the third neck crease extending from right mastoid process to the contralateral mastoid process overlying the hyoid bone. (figure 6)



Figure 6: - Visor Incision Marking



Subplatymal flap was raised till the inferior border of the mandible while preserving marginal mandibular nerve. Tumor was identified and delineated. Surgical template was adapted and contoured to the original shape of the mandible prior to resection.

Segmental mandibulectomy was performed from 35 to 45 region using bur in hand piece under copious saline irrigation to achieve a three dimensional resection of the lesion. (figure 7 & 8)

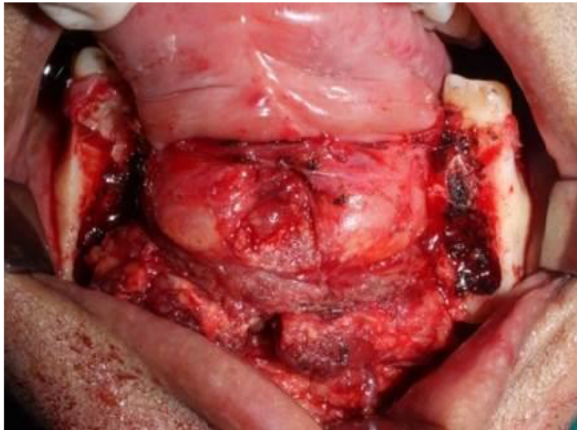


Figure 7: - Intraoperative view of surgical field post tumor resection



Figure 8: - Resected Specimen

Fibular microvascular free flap was harvested in its classical technique with the desired muscular component over peroneal vascular pedicle. Microvascular anastomosis carried out using 8-0 ethilon in the following manner:-

- Peroneal artery with Facial artery.
- Peroneal vein with Internal jugular vein.

The graft was osteotomized and recontoured to simulate the shape of native mandible and secured to the remaining part of mandible using a 2.3mm titanium reconstruction plate and 2.3mm x 8mm titanium screws. The soft tissue component of the flap was secured to the surrounding tissues using 3-0 vicryl. (figure 9)



Figure 9: - Recontoured fibula secured to the defect site using titanium miniplates and mini screws

After achieving haemostasis layered primary closure of the neck wound and donor site wound was accomplished using 3-0 vicryl and skin stapler. (figure 10)



Figure 10: - Primary closure of the surgical wound

The surgical specimen was sent for histopathological examination which revealed cavernous hemangioma of anterior mandible and adjacent soft tissues.



Figure 11 : - Post operative view after 6 months  
a: - Extra oral frontal view  
b: - Intraoral view showing well healed surgical site

## Discussion

Hemangiomas in an intraosseous location are rarely reported entities, especially in the jaws. When found, the majority are present during the second decade of life. These lesions have been considered by the World Health Organization as a true benign vasoformative neoplasms, or a developmental condition of endothelial origin<sup>9</sup> whereas, many authors understand them to be a hamartoma originating from the proliferation of mesodermal cells that undergo endothelial differentiation, become canalized, then vascularized.<sup>10</sup>

Clinically, the patient may be completely free of symptoms or may present with pain of variable intensity, soft tissue swelling, bluish color in the mucosa, tooth mobility, displacement of the mandibular canal, expansion of the cortices, deformity of the arch shape, resorption of roots, ageneses or accelerated dental exfoliation.<sup>4,6,7</sup>

Intraosseous hemangiomas possess a diverse radiographic appearance and thus cannot be precisely diagnosed on plain films. In general, they present with an osteolytic pattern possessing a multi-locular “soap bubble” appearance with irregular, poorly defined margins.<sup>1,2,4,7,8,10</sup>

Many pathologic entities may be described in similar radiographic patterns and a differential diagnosis could include giant cell lesion, odontogenic tumor, aneurysmal bone cyst, hemangioma and odontogenic myxoma. As such, angiography plays a crucial role in diagnosis as it can confirm the suspicion of the vascular lesion, delineate its margins, and indicate feeder vessels.<sup>11</sup> Definitive diagnosis of an intraosseous hemangioma cannot be made without histological examination, but due to the risk of severe hemorrhage, needle aspiration should precede biopsy of any such suspicious lesion. The presence of easily aspirated blood with significant volume and brisk hemorrhage from the puncture site should warrant suspicion.<sup>12</sup>

Histologically, intraosseous hemangioma are masses of endothelial cells that form vascular spaces of varying sizes mixed in a stroma of connective and fibrous tissue with visible trabecular bone.<sup>10</sup>

They can be divided into three groups according to the size of the vascular spaces: cavernous, capillary and mixed. The cavernous, the most common, are composed of larger vessels and spaces covered by endothelium and scant stroma, capillaries are small vessels with pronounced cellularity and stroma. Mixed lesions show both capillary and cavernous characteristics. All present a proliferative stage characterized by rapid growth through the first years of life, with prevalence in the vascularization, increase activity of endothelial cells and mast cells, followed by a regressive phase characterized by an increase in the fibroblastic matrix and a decrease of the vascular component.<sup>1,3</sup>

In the presence of suggestive clinical signs and symptoms, radiographic appearance, positive aspiration, and arteriogram, biopsy may be deferred and treatment should

be initiated with a presumptive diagnosis of intraosseous hemangioma.<sup>2</sup>

A number of treatment modalities have been recommended and the selection is largely dependent on the size of the lesion, the age of the patient, and any anticipated complications. Suggested modalities of treatment include surgery, sclerotherapy, cryotherapy, embolization and irradiation.

Surgery is widely accepted as treatment of choice for central hemangioma of the mandible; several techniques are described in the literature.<sup>11,12,20</sup> Curettage performed after removal of the buccal cortical plate to expose the lesion, followed by replacement of the buccal plate after removal of the lesion, has been reported. This manoeuvre preserves the continuity of the mandible.<sup>21,45</sup> Many authors have reported that simple curettage leads to uncontrollable bleeding and incomplete removal of the lesion which leads to recurrence.<sup>11,12,13,14,20</sup>

En-block resection of the lesion with or without immediate bone graft reconstruction is considered to be the preferred surgical procedure.<sup>6</sup> By placing the resection lines in uninvolved tissue, one can avoid manipulating the lesion and thereby reduce the risk of massive hemorrhage while ensuring a three dimensional resection of the lesion thereby minimizing the chances of recurrence. Regardless of surgical technique, most cases involved ligation of feeder vessels before removal of the lesion. In many cases the ipsilateral external carotid artery was ligated whereas in other cases bilateral ligation of the external carotid arteries was performed.<sup>12,20</sup> Many times, this has been effective in reducing intraoperative hemorrhage; however, in several case reports ligation of major vessels did not significantly reduce hemorrhage.<sup>11,12,26</sup> This reinforces the importance of angiography to identify anomalous feeding vessels and therefore guide preoperative planning. Surgical removal should be done immediately after ligation or embolization of major vessels because collateral vessels are known to develop quickly.<sup>13,24,26</sup>

Sclerosing agents such as sodium morrhuate, sodium psylliate, and absolute ethanol have been injected into these lesions.<sup>16-19</sup> These agents induce an inflammatory reaction within the endothelium that ultimately results in fibrosis and obliteration of the vessels. Sclerosing agents have been used primarily in soft tissue hemangiomas, although successfully treated cases of central hemangioma of the mandible have been reported. Injection of the sclerosing agent limits the destruction to the endothelium without harming the bone.<sup>20</sup> Sclerosing agents produces long-term arterial thrombosis in large arteries with eventual replacement by connective tissue and is not recommended for the management of larger lesions.<sup>18</sup>

Cryotherapy for treatment of small cavernous hemangioma has also been reported.<sup>21</sup> However, cryotherapy tends to damage the lesion, its surrounding tissue, such as adjacent nerves, and viable bone. Although, its merits include ease of application, cost effectiveness and ability to produce



lesser postoperative scar tissue formation.<sup>17</sup> Hence cryotherapy should be reserved for small lesions and those detected on table, without aspiration or CT Scans.

Embolization of large vessels feeding the lesion is another reported treatment option.<sup>22,23</sup> Azzolini *et al* introduced a novel technique of intraoral treatment by extracting the teeth and then cleaning the underlying cavity through the alveolar process and packing the mandible with oxidized cellulose after super selective intra-arterial embolization (SIAE). This technique offered the advantages of preserving the bone tissue, dental follicle, erupted tooth, and inferior alveolar neurovascular bundle. Under fluoroscopic control a catheter is inserted into the appropriate vessel and the material delivered.<sup>23</sup> Various embolizing agents include silicone pellets, gelatin sponge, muscle, lead pellets,<sup>12,24</sup> or isobutyl cyanoacrylate.<sup>17</sup> This procedure is technically difficult and carries complications, notably embolization of pulmonary or cerebral vessels, allergic reactions, avascular necrosis of bone, delayed root development, defective mandibular growth.<sup>23,24</sup> Furthermore, embolization of feeder vessels may shrink the lesion only temporarily until collateral vessels develop.<sup>22</sup> Therefore embolization has been used in conjunction with surgery to reduce intraoperative bleeding.<sup>20,22,23</sup>

Radiotherapy is also considered as an alternative to conventional treatment options due to reduction in the total tumoral volume and malignization of the lesion. But, owing to potential side-effects such as damage to the adjacent soft tissues, residual scarring and malignant transformations renders it as an unacceptable treatment modality for the management of hemangioma of maxillofacial region.<sup>14</sup>

The treatment of choice is ligation of the feeding vessels followed by radical resection of the tumor and primary reconstruction. Post-surgery, these lesions have a good prognosis in terms of recurrence and the majority of these patients enjoy a good quality of life provided, careful reconstruction has been undertaken to restore optimal form and function.<sup>4,5</sup>

Even with proper identification and pre-operative planning, significant bleeding is to be expected intra-operatively, and clinicians should be prepared for rapid transfusion.<sup>23-28</sup> Upon successful surgical removal of the lesion, appropriate reconstruction measures may be taken to restore the patient to optimum form and function.<sup>28</sup>

### Conclusion

Clinicians should be well aware of the possibility of an intraosseous hemangioma in patients who present with a bony expansile lesions pertaining to the maxillofacial region. Failure of consideration of this dilemmatic lesion as a differential diagnosis could lead to significant blood loss eventually beholding a life threatening condition. The management of a patient with an intra-osseous hemangioma depends on many variables such as age of the patient, extent of lesion, involvement of vital structures and affordability to treatment. Considering all these aspects, one

should tailor make the treatment plan suitable to each individual.

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