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Original Research Paper

Dentistry

INTRAOSSEOUS CAVERNOUS HEMANGIOMA OF MANDIBLE : A RARE CASE REPORT

Ajay Kumar*	PG Resident, Dept. of OMFS, HDC&H, *Corresponding Author
Rajesh Dhirawani	Professor Emeritus, Dept. of OMFS, HDC&H
Sanyog Pathak	Professor & HOD, Dept. of OMFS, HDC&H
Sumit Asrani	Reader, Dept. of OMFS, HDC&H
Ankit Sharma	Consultant, JH&RC
Sugam Neema	PG Resident, Dept. of OMFS, HDC&H

ABSTRACT Intraosseous hemangiomas, mainly of the jaw bones are quite uncommon lesions (0.5-1%) of all intraosseous tumors. They are most commonly seen in second decade of life with a higher female predilection (2:1). The most common sites of occurrence are skull and vertebral column and rarely seen in the mandible. The origin of hemangiomas is still debatable. Hemangiomas are rare benign vasoformative neoplasms of endothelial origin. They usually present as a firm, painless swelling of the jaw bone with or without facial asymmetry. Cavernous hemangioma poses a diagnostic dilemma and mimics lesions like ameloblastoma, central giant cell granuloma, osteosarcoma, fibrous dysplasia . Here we are presenting a case report of a 28 year old female with intraosseous cavernous hemangioma of right body of mandible which was treated surgically and followed by reconstruction.

KEYWORDS : Cavernous hemangioma, intraosseous, mandible.

INTRODUCTION

Intraosseous hemangioma is a benign, slow growing neoplasm that consists 1% of all benign tumors arising from the bones. Hemangiomas mostly arise from the soft tissues, and intraosseous hemangiomas are uncommon.[1] Hemangiomas in the facial skeleton are rare; when they occur, two thirds are found in the mandible. Statistically they occur more often in females, by a 2: 1 ratio, and manifest during the second decade of life, although cases have been reported in a wide range of age groups.[2] The origin of cavernous hemangioma is debatable. According to Shira and Guernsey, it is a true benign neoplasm formed because of endothelial proliferation which differentiates into blood vessels. Some authors suggest it to be a hamartomatous lesion which develops from the proliferation of mesoderm that further undergoes endothelial differentiation which is later on localized and vascularised.[3,4]

The diagnosis of this lesion can be difficult because neither the history nor the clinical findings are sufficient to suspect an intraosseous cavernous hemangioma.[5,6] Mostly asymptomatic but may present with symptoms such as bluish slow growing mass, teeth mobility, pulsatile sensation, compression of surrounding structures, haemorrhage and overall altered quality of life.[7]

Orthopantomogram (OPG), Computed tomography (CT), Magnetic resonance imaging (MRI), and CT angiography are useful for diagnostic purpose. Central hemangiomas of bone arise from vessels within the marrow spaces and may comprise arterial and venous vessels. Microscopically, engorged vascular sinuses are present with an endothelial lining supported by a connective tissue stroma interspersed by bony trabeculae, which are usually arranged at right angles to the surface.[8]

The radiographic presentation of the cavernous hemangioma is nonspecific. The lesion appears as a radiolucency that could have unilocular or multilocular, reticulated, honeycomb, or sunburst appearance when viewed tangentially. The correct diagnosis of cavernous hemangioma is relatively challenging due to similar radiologic features of ameloblastoma, odontogenic myxoma, fibrous dysplasia, and aneurysmal bone cyst.[9] In this paper, we presented a rare case report of intraosseous cavernous hemangioma in the mandible , diagnosed and managed surgically at our centre.

CASE REPORT

A 28 year female reported to our centre with the chief complaint of pain and swelling in the right side of her lower jaw since 5 years. The history of present illness revealed that she first noticed the swelling 5 years back in her right lower jaw which was associated with dull and intermittent pain. The patient was operated for the same under local anaesthesia 3 years back at some other centre. The chief complaint of the patient persisted even after the operation and the swelling gradually increased to the present size and was associated with dull and continuous pain. There was no history of pus discharge and no aggravating or relieving factors were present. No relevant medical history and history of spontaneous bleeding or paresthesia was given.

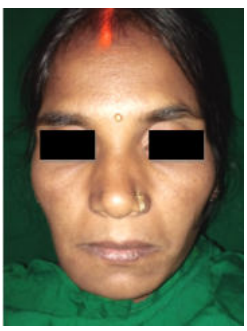

Figure 1 – Frontal view of the patient

On extra-oral examination, facial asymmetry was present on the right side of face and the swelling of size approximately 3cm × 3cm was extending from midline to the middle of the body of mandible. The swelling was superiorly extending from right angle of mouth to right angle of mandible inferiorly. A scar was present on the right side in submandibular region. Right nasolabial fold was partially obliterated. On palpation, the swelling was bony hard, tender, smooth, diffuse with raised temperature. Right submandibular lymph node

palpable, tender, non-fixed, two in number of size measuring about 0.7*0.6cm & 0.5*0.6cm.

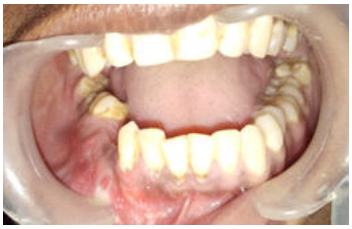

Figure 2 - Intraoral picture of lesion

Intra-oral examination revealed a bony hard swelling present in the region of 44,45,46 with obliteration of buccal vestibule. There was expansion of buccal and lingual cortical plates.

Supra eruption of 46 present and 44, 45 were missing. No mobility was present with any teeth.

Orthopantomograph revealed multilocular radiolucency with numerous radiopaque trabeculations extending from 32 to right mandibular angle region involving ramus giving a honeycomb appearance. 46 was seen floating in the bone in OPG. CBCT imaging confirmed the radiographic findings.

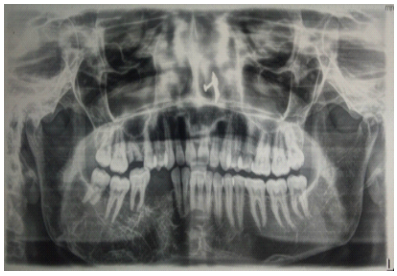

Figure 3 - Orthopantomograph showing multilocular radiolucency with numerous radiopaque trabeculations

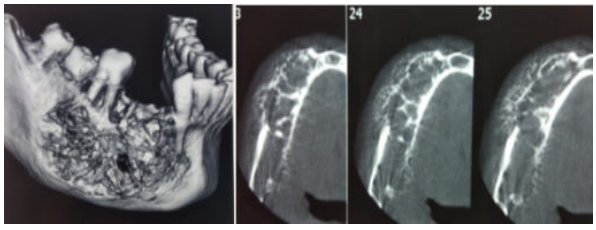

Figure 4 – CBCT showing involvement of right body of mandible

CBCT scan findings revealed an ill-defined, heterogenous, multiloculated, and multiseptated osteolytic lesion involving right mandibular body and extending till the ramus.

Surgery was planned, under general anesthesia, segmental resection of mandible was done from 34 to sigmoid notch of right side of mandible followed by reconstruction of the defect with reconstruction plate. The excised specimen was sent for histopathological diagnosis, which confirmed the diagnosis of cavernous hemangioma.

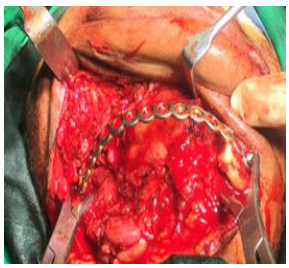

Figure 5 – Intraoperative picture of defect

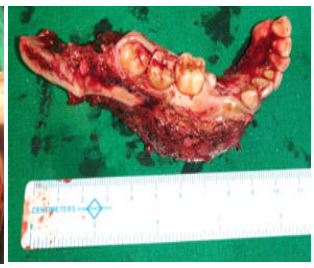

Figure 6 – Resected specimen

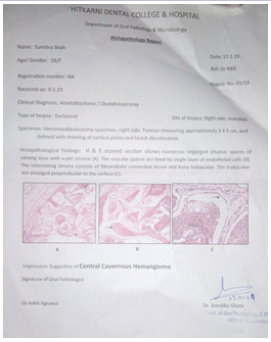

Figure 7 – Biopsy report

DISCUSSION

Hemangiomas especially of the jaws, are rarely reported pathologies. The majority of lesion is seen during the second decade of life and mostly in the mandible with a female to male ratio of 2:1. World Health Organization have considered these lesions as true benign vasoformative neoplasms, or a developmental condition of endothelial origin whereas, some authors believe them to be a hamartoma derived from the proliferation of mesodermal cells that undergo endothelial differentiation, become canalized and then vascularised.[10]

There are two variants of hemangioma: Peripheral and central. Peripheral hemangioma is originated in the periostic vessels that grow into the medullar bone, while central hemangioma is originated into the medullar bone and grow toward the cortical bone. Histologically, hemangioma can be divided into three groups: Cavernous (the most frequent one and is located into the mandible), capillary and mixed.[11]

The lesion occurs as firm, painless bony swelling which may cause gross facial asymmetry but mostly they are asymptomatic. Pain and paresthesia are not characteristic features but may be present. Intraorally, common findings are obliteration of vestibule, expansion of buccal or lingual cortex, mobility, and displacement of teeth. Occasionally, root resorption also seen. Supra-eruption, premature exfoliation of primary teeth, and early eruption of permanent teeth also have been seen.[12]

Intraosseous hemangiomas shows a varied radiographic appearance and thus cannot be accurately diagnosed on plain films and can simulate many other lesions of the jaw. The lesion can present with an osteolytic pattern having a multilocular "soap bubble" appearance with irregular, poorly defined margins.[10]

There are different treatment modalities present for cavernous hemangioma but surgical intervention is generally accepted as the definitive treatment, with en bloc resection is the recommended procedure. Other treatment modalities include embolization, intralesional corticosteroids or sclerosing agent injections and radiotherapy. Surgical excision with reconstruction of the defect remains the preferred treatment modality. The prognosis after complete excision is excellent and recurrence is normally rare.

CONCLUSION

The diagnosis of facial bone hemangiomas is somewhat difficult to make, because the lesion is usually asymptomatic. The early diagnosis of intraosseous hemangioma is essential both for preventing uncontrollable haemorrhage and even death during biopsy or surgery. The propensity for anatomic variation emphasizes the importance of angiography for proper identification of vessels associated with the primary lesion. The treatment modality should be carefully planned as per the patient's age, clinical features, extent of the lesion, and systemic medical status.

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