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Case Report

Cavernous Hemangioma: A Rare Case Report

Dr. K Saraswathi Gopal[1], Dr. J K Singh Kshatri[2], Dr. K Ramesh Kumar[3]
[1]HOD, Dept of oral medicine and radiology, Meenakshi ammal dental college, Chennai.
[2]PG Student, Dept of oral medicine and radiology, Meenakshi ammal dental college, Chennai.
[3]Assistant Professor, Govt Dental college, Vijayawada, AP.

***Corresponding author**
Dr. K Saraswathi Gopal
Email: drjksinghk@gmail.com

Abstract: Intraosseous hemangiomas are the rarest lesion of jaw bones (0.5–1%) occurs more commonly in vertebral column, skull bone, and rarely in mandible. Mostly occurs in the 2nddecade of life with female: male predilection (2:1). Origin of hemangiomas is still not clear. World Health Organization considers it as a true benign neoplasm of vascular origin, and many authors believe it to be a hamartoma. Cavernous hemangioma is very difficult to diagnose due to variable clinical and radiological features. Management is very difficult because of massive vascular network in that region. Here we are presenting a case report of 64 years old female with cavernous hemangioma of anterior mandible, which was treated with surgical resection of mandible and followed by reconstruction.
Keywords: Hemangioma, Cavernous hemangioma, intra osseous, Mandible, vascular origin, benign neoplasm

INTRODUCTION:

Hemangioma is a benign tumor that is characterized by the proliferation of blood vessels and these are usually found in soft tissues [1]. The intraosseous hemangiomas or cavernous hemangiomas are relatively rare and constitutes about 0.5-1 % of all intra osseous tumors [2]. When osseous components are involved the common sites of involvement include vertebral column and skull bones jaw involvement is seldom and are described as rare benign vasoformative neoplasm of endothelial origin [2].

Majority of these jaw lesions are found to occur during second decade of life with mandible as common site of occurrence. Female to male ratio is 2:1 [3]. It is also believed to be a hamartoma that arises due to proliferation of mesodermal cells which undergo endothelial differentiation become canalized and vascularized in the mandible the lesion is commonly found in the body region although few condylar tumors have also been reported[3]. 65% of the tumors were reported in the molar and premolar region

Intraosseous hemangiomas of the jaws present as a firm painless swelling of the jaw bone with or without facial asymmetry [4]. Usually it is asymptomatic but may present as a slow growing blueish mass resulting in pulsations, dearranged dentition, tooth mobility, recurrent bleeding due to trauma, paresthesia and discomfort. It may also result in compression of surrounding structures[4]. Orthopantamogram (OPG), computed tomography and MRI remain the principle diagnostic modalities.

CASE REPORT:

A 64 year old female reported to department of oral medicine and radiology with the chief complaint of swelling on the lower face for the past 3 year. History of present illness revealed trauma before 3 years, initially the swelling was bigger in size which has decreased to attain the present size and shape. Swelling was associated with non radiating dull pain. After 1 year, swelling size became stagnant with a lower intensity of pain. At present, she experiences a hard swelling which is painful on the application of pressure. No history of spontaneous bleeding or paresthesia in that region. No relevant medical or surgical history.

Extra oral examination revealed asymmetry on the right lower third of the face. A single well defined prominent swelling on the right chin region was approximately 6 cm × 8 cm in size elevating the lower lip extending from the inferior border of mandible to superiorly to the vermelion border anteriorly crossing the midline to about 2cm towards left and posteriorly 3cms beyond the right commissure of the lip. On palpation a bony hard swelling with tenderness was present (Figure 1 & 2).

Mouth opening was normal with grade 3 mobilty of 31, 32, 41, 42, 43, 44 and displacement of lower anteriors towards lingually. There was diffuse swelling obliterating the buccal vestibule, extending from 31 to 45 region with labial cortical expansion was present with blanching of mucosa without any pulsations observed and crater like ulcer measuring approx. 2x0.5 cms with smooth margins is seen on the surface of the swelling due to impingement of upper incisors (Figure 3). Based on clinical features a provisional diagnosis of benign bony lesion is given.

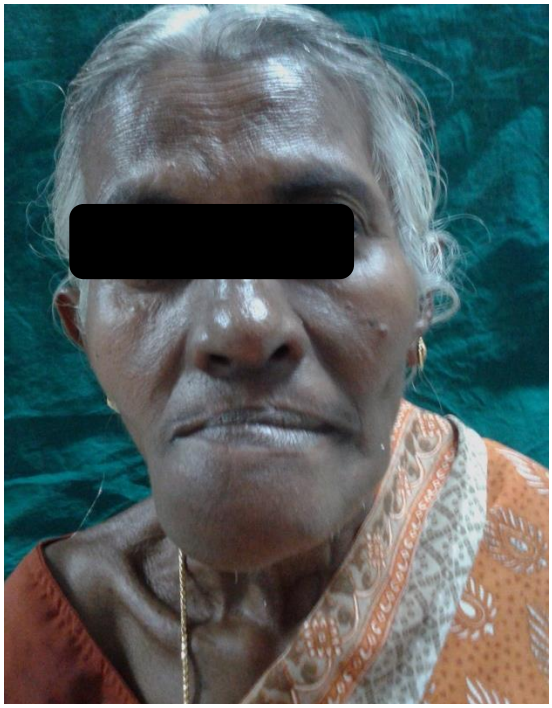

Fig 1: Extra Oral Picture showing the facial asymmetry

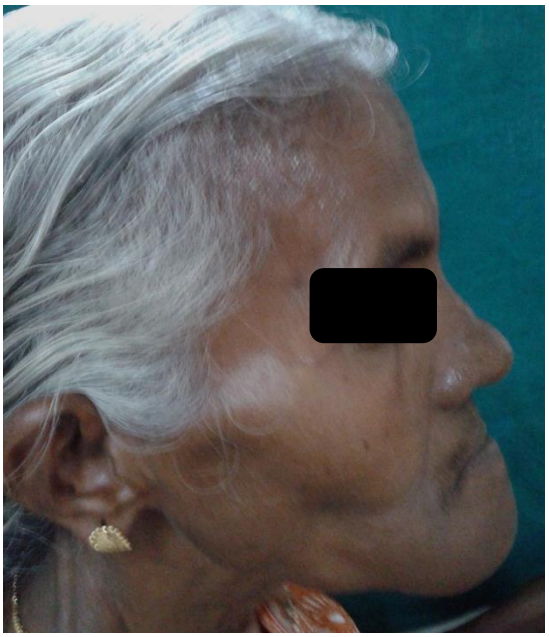

Fig 2: Lateral View

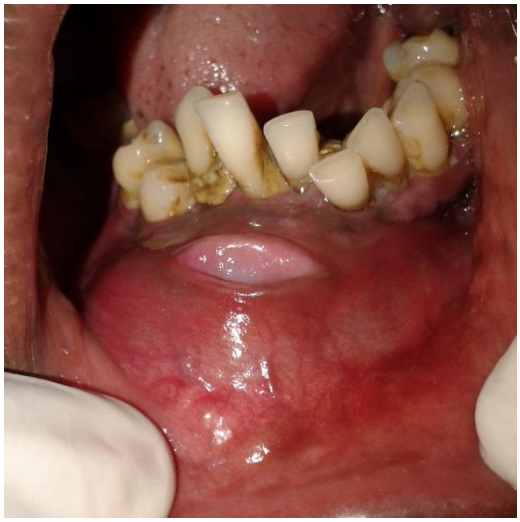

Fig 3: The intra oral picture showing the swelling obliterating the buccal vestibule with crater like ulceration

Investigations:

The occlusal radiograph revealed a mixed radiopaque and radiolucent lesion evident in the anterior mandible measuring approx. 5x5 cms extending from 34 to 46 regions with expansion of buccal and lingual cortical plates (Figure-4).

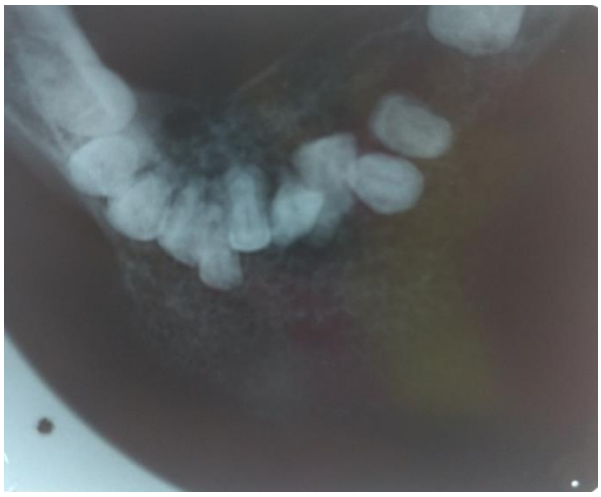

Fig 4: Occlusal Radiograph of anterior mandible shows coarse trabecular pattern

OPG revealed a single ill defined, non-corticated mixed radiolucent and radio opaque lesion evident in the anterior mandible measuring approx. 5x5 cms with multilocular appearance surrounded by coarse, dense and well defined trabeculae and the normal path of the inferior alveolar canal is altered in to a serpiginous shape on the right side. Generalized moderate to severe crestal bone loss observed, root stump in relation to 26 and 35.

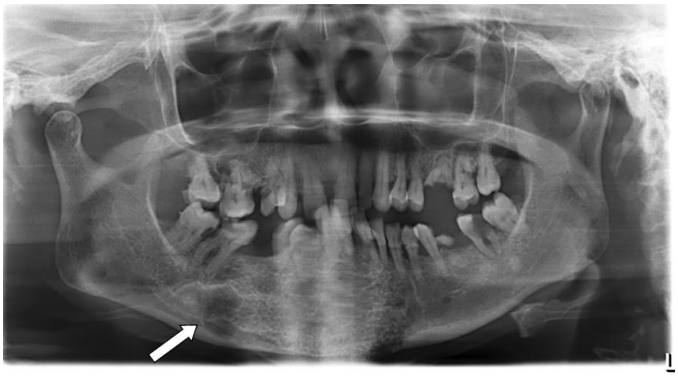

Fig 5: OPG revealed a single ill defined, non-corticated mixed radiolucent and radio opaque lesion evident in the anterior mandible with multilocular appearance surrounded by coarse, dense and well defined trabeculae

The axial view of CBCT of mandible revealed an expansile lesion evident in relation to anterior mandible leading to the expansion of the buccal cortical plate with mixed hyper and hypodense internal structure (Figure-6). Suggestive of osteosarcoma, fibrous dysplasia and central gaint cell granuloma.

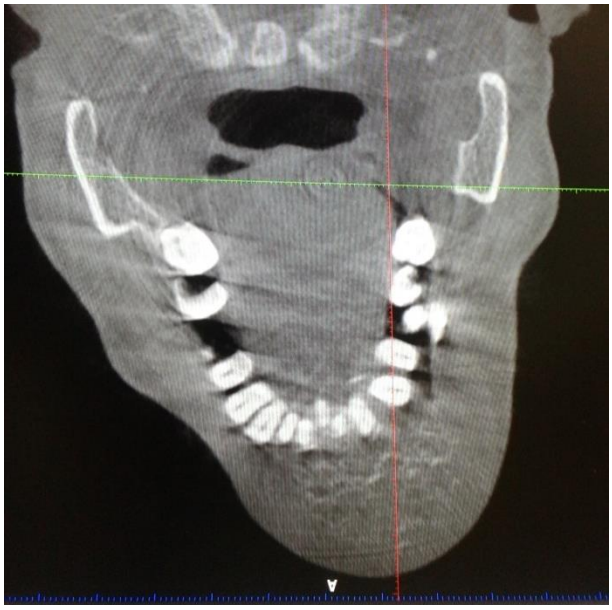

Fig 6: The axial view of CBCT of mandible revealed an expansile lesion evident in relation to anterior mandible

Treatment:

Surgical treatment was planned and hemi mandibulectomy was performed and the specimen was sent for histopathological examination which showed numerous dilated vascular spaces lined by endothelial cells and filled with RBCs and intervening trabeculae of bone is seen suggestive of a vascular lesion (Figure 7).

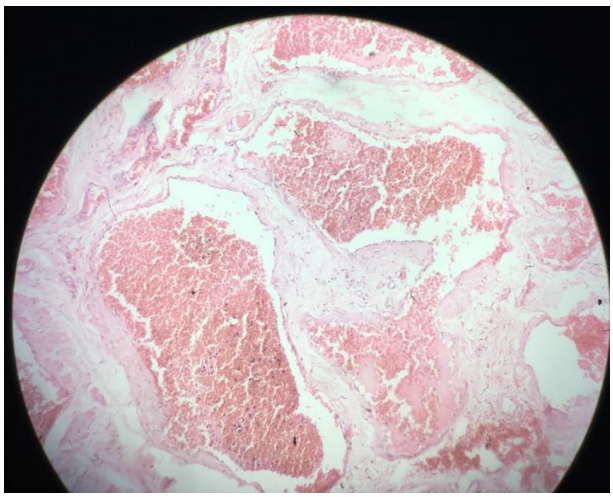

Fig 7: Histopathological showed numerous dilated vascular spaces lined by endothelial cells and filled with RBCs and intervening trabeculae of bone.

Based on clinical, radiological and histopathology reports a final diagnosis of cavernous hemangioma was made.

DISCUSSION:

Intra osseous hemangiomas are rare benign vasoformative neoplasm of endothelial orign [4]. The initial endothelial proliferation resulting in a true benign neoplasm explains the origin of cavernous hemangioma and was described by Shira and Gueenscy(1965) it was also believed to be a hamartoma that results from proliferation of mesoderm which undergoes endothelial differentiation subsequently canalized and vascularized [1, 3]. Intraosseous variant of hemangiomas seldom involve the jaw bones when compared to the vertebrae and skull. Mandible is commonly affected

predominantly in molar and premolar regions and condylar tumors were also reported [5].

Radiographic features of cavernous hemangioma simulate many other bony lesions though these features are pathognomic of the tumor [1]. Several variations in the radiographic appearance have been described by various authors which are to be considered in the differential diagnosis of several other tumors and cysts of bone [6]. Presence of a parallel or tube like arrangement of radiopaque striae serves as an indicator for hemangioma according to Langland *et al.;* The trabecular arrangement as that of the spokes of a wheel has been as described by worth which radiates from center to the periphery. Classical sunray or sunburst appearances have also been proposed by many authors where in the trabeculae are coarse perpendiculars to bony surface. The periphery of the lesion can either show a well-defined or ill-defined corticated area with scalloped margins. The soap bubble or honey comb appearance may result from patchy, multicystic osteolytic areas. These tumors may also sometimes exhibit elements of fibrous connective tissue, osseous tissue and cavernous blood vessels [1].

The following lesions and tumors are to be considered in the differential diagnosis Osteosarcoma, Fibrous dysplasia, Central giant cell granuloma, Ameloblastoma, Multiple myeloma when skull radiographs exhibit punched out radiolucencies [3].

Histology of hemangioma lesions

The cavernous form of hemangioma consists of large dilated blood vessels with thin walls, each showing an endothelial lining. These sinusoidal spaces are usually filled with blood, although an admixture with occassional lymphatic vessels occurs in some instances [7].

On the basis of histology, hemangioma has been classified into (1) capillary, (2) cavernous, and (3) mixed variant. 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 describes the development of hemangioma in three stages (1) early: Highly vascular, (2) intermediate: Exhibits blood clotting, and (3) terminal: Various stages of ossification [2]. Variety of syndromes consists hemangiomas as part that includes Osler Weber Rendu Syndrome, Sturge Weber Dimitri Syndrome, Kesabach Merritt Syndrome, Maffuci Syndrome, Von Hippal Lindau Syndrome, Kippel Trenaunay Weber Syndrome [4].

Several treatment modalities are recommended and the choice of treatment depends on the size of the lesion, the age of the patient and suspected complications. Cases which do not show remission or those which arise in older persons have been treated in variety of ways including [8].

1. Surgery
2. Radiation therapy
3. Sclerosing agents such as Sodium Morrhuate or Psylliate, injected in to the lesion
4. Carbon dioxide smoke
5. Cryotherapy and
6. Compression

Intralesional corticosteroids have been used successfully to reduce the size of the lesion but may result in ulceration [9]. Prognosis of cavernous hemangioma is excellent since it does not become malignant or recur after adequate removal or destruction.

CONCLUSION:

The hemangioma is a benign proliferation of endothelial cells common in the head, neck and relatively rare in the oral cavity. 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. Treatment modality should be carefully planned based upon patient's age, clinical features, extent of the lesion, and systemic medical status.

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