CASE REPORT

Hemangioma: A rarest entity
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Abstract
Hemangiomas are benign congenital lesion which has endothelial origin, but still evidence of its origin is in dilemma as few writers consider it as a true neoplasm and on the other hand as a hamartomatous lesion. The frequency of central hemangioma in jaws is less. Clinically, the patient may be asymptomatic or there may be the presence of pulsatile bleeding, slow growing bluish mass, mobile teeth, and deranged dentition, early dental exfoliation and discomfort in normal life hence on clinical basis the mucosal and soft tissue lesions can be easily diagnosed, but intrabony lesions are difficult to discriminate. Its clinical and radiographic features are comprehensive therefore, a suitable diagnosis is mandatory. Treating central hemangioma is very difficult because of its profuse vascularity. Out of various remedies, available surgery is being most frequently used. In this article, we report a 22-year-old male patient with central hemangioma located in right body of the mandible with ambiguous clinical features, radiographical, and histopathological features.

Keywords
Hemangioma, mandible, vascularity

Introduction
Hemangioma is the utmost most common benign inherited lesions in individuals which are congenitally present or appear soon after birth, and are known by the proliferation of blood vessels.[1-4] As stated by Shira and Guernsey, it is a true benign neoplasm and its occurrence is due to endothelial proliferation which differentiates into blood vessels whereas some authors advocated it to be a hamartomatous lesion which arises due to mesodermal proliferation that endures endothelial differentiation which is supplementarily localized and vascularized.[5,6] According to Thoma, hemangioma has been histologically classified into two forms first as peripheral form (originating from periosteum) and second as central or intraosseous form (originating from central spongiosa).[4] There are two central types, cavernous, the most common and infrequent capillary type. Capillary hemangioma comprises a solitary coating of endothelial cells enclosing many smaller capillaries buttressed in a connective tissue stroma of changeable concentration, whereas cavernous hemangiomas comprise the solitary coating of endothelium enclosing large, fragile vessels or sinusoids detached by thin septa connective tissue septas.[7] The prevalence of central (intraosseous) type is more in vertebrae and skull and less in jaws with a rate of <1% of all intraosseous tumors.[8,9] It is more prevalent in females than males at the ultimate incidence between the second and fifth spans of lifetime.[10,11] Cavernous hemangioma is mostly of congenital origin occurring frequently in mandible compared to maxilla and nasal bones.[12] In mandible, the body region is mostly affected, whereas some condylar tumors have also been reported.[13] Usually, it is asymptomatic but sometimes patient may complain of discomfort, pulsation, bluish slow growing mass, compression of surrounding structures, mobile teeth, and hemorrhage.[11] Orthopantomogram (OPG), computed tomography, magnetic resonance imaging, ultrasonography (USG) are the most common diagnostic modalities used.

In this article, we reported a rare presentation of case diagnosed as central hemangioma in right mandibular body.

Case Report
A 22-year-old male reported with the chief complaint of swelling on the right lower third of the face since 1-year. History of present illness revealed an asymptomatic swelling present on the right lower third of the face which gradually increased in shape and size and reached to its present size around 6 months back. Swelling was associated with nonradiating dull pain with no history of spontaneous bleeding or paresthesia.

No relevant medical history was reported. Extraoral examination revealed the presence of facial asymmetry on lower third right of the face with a diffused swelling approximately 4 cm × 3 cm in size and present over the right body of the
mandible, extending superoinferiorly 2 cm below ala-tragus line to 1 cm below inferior border of the mandible, and anteroposteriorly 1 cm away from the corner of the mouth to 1 cm away from the angle of the mandible with smooth surface and without any discoloration on comparing to the other normal side. On palpation, bony hard, smooth, tender, and diffuse swelling was present in the lateral aspect of the right mandibular body involving the inferior border without any rise in temperature.

His mouth opening was normal with acceptable occlusion; mandibular right second molar was in hypo-occlusion. In the concerned area teeth present were vital and firm [Figures 1 and 2]. There was diffuse hard swelling obliterating the buccal vestibule, extending from mandibular right first premolar to retromolar region [Figures 3 and 4].

Radiographic analysis was performed on the basis of OPG and USG face. OPG exhibited coarse trabeculation, haziness, and increased radio-opaque striations from center to periphery extending from the first premolar to molar region [Figure 5].

USG was done which revealed 3.4 cm × 1.7 cm × 1.8 cm sized anechoic lesion with internal septations noted in the right submandibular region anteromedial to the right submandibular gland. Right facial artery is seen traversing through the lesion and 2.7 cm × 1.2 cm sized similar lesion noted over the body of right mandibular anterior to right masseter muscle without evident communication with the right submandibular lesion. Slow flow was evident on the color Doppler study.

After fine needle aspiration cytology, which yields approximately 6 ml of straw colored fluid, and cytological report came as central hemangioma of right body of the mandible [Figure 6].

**Discussion**

In maxillofacial region, the vascular lesions incorporate a wide range of lesions. These vascular lesions were classified for the first time by Mulliken and Glowacki in 1982 on the basis of histological characteristics, as either hemangiomas or
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vascular malformations. The utmost common cutaneous tumor of infancy is hemangioma occurring in up to 3-5% of all children and revealed rapid development which is further followed by an involution which is slow and spontaneous or regression within 5-7 years. Contrastingly in hemangiomas, vascular malformations enlarge consistently as child grows and do not undergo spontaneous involution. The striking feature of hemangioma, a benign tumor is blood vessel proliferation of endothelial origin. The term “hemangioma” is derived from the Greek term hema denoting “blood,” angio denoting “vessel,” and the suffix-oma denoting “tumor.” A hemangioma which is described as blood vessels proliferation producing a neoplastic mass. The prevalence of in site of occurrence in central (intraosseous) type is more in vertebras and skull as compared to jaws. Frequently cavernous type (large-calibre vessels) of central hemangioma occurs than capillary type (small caliber vessels). Central hemangioma can be either asymptomatic/symptomatic presenting signs and symptoms as uneasiness, discharging blood from marginal gingiva of the teeth surrounding region of the lesion, bluish discoloration of gingiva and mobile teeth. In the reported case, the presence of facial asymmetry due to extraoral facial swelling on right lower third region. Intraorally, central hemangiomas present as painless swellings pertaining to premolar-molar region.

Radiographically, there is no distinguishable appearance and can pretend many other bony lesions. Lesions are less frequent unilocular radiolucent and more frequent is multilocular radiolucent with small loculations depicting honeycomb appearance or large loculations depicting soap-bubble appearance. Worth has added one more striking feature where trabeculae resemble the spokes of a wheel, radiating from the center of the lesion. The appearance of central hemangiomas in radiographs as well-defined margins, wherein the with enlarged marrow spaces surrounded by uneven, condensed, and well-defined radiopaque trabeculae. However, in the present reported case OPG, revealed larger radiolucent areas with coarse trabeculae giving it a multilocular radiolucency. Radiographically, a differential diagnosis of ameloblastoma, cavernous hemangioma, giant cell lesion, cyst, and myxoma could be given.

Clinical history, examination findings, radiographs, and scanning examination illustrated many features which showed characteristic of central hemangioma.

For avoiding complications as uncontrollable hemorrhage and even death during biopsy or surgery, therefore for its early diagnosis dental practitioners awareness about this lesion clinically as well as radiographically is mandatory.

There is high risk of bleeding so incisional biopsy is legally contraindicated.

There are various treatment modalities available which usually depends on the complexity of the lesion which includes steroid therapy, sclerosing agents, irradiation, and surgical excision with or without ligation of vessels, embolization, laser therapy, and replacement of resected area with iliac bone graft. We should plan treatment only in some conditions where esthetics is concerned or in case of very large size or unusual location of the lesion to avoid uncontrolled bleeding.

Conclusion

In maxillofacial region, intraosseous vascular lesion is the rarest entity and can be challenging for diagnosing various jaw lesions. Treatment planning is a challenge and should be based on patient’s age, clinical features, extent of the lesion, and systemic medical status otherwise it can lead to various life-threatening complications. Hemangiomas must be undertaken as a differentiating feature so that proper safety measures should be taken into consideration before any surgical treatment is commenced.

Figure 5: Cropped image of orthopantomograph of the patient depicting multilocular radiolucency on right side

Figure 6: Fine needle aspiration cytology was done showing straw colored fluid
References
