Hemangioma was previously defined as the variety of developmental vascular anomalies. However, in recent times, they are considered to be benign tumors of infancy characterized by a rapid growth phase with endothelial cell proliferation, followed by gradual involution. Hemangioma is mainly located in the soft tissues. Intraosseous hemangioma constitutes less than 1% of the reported cases of hemangiomas. They mainly occur in the second decade of life especially in women. The most common location is the vertebral column and skull, while the maxilla or mandible is a quite rare location. The origin of central hemangioma is debatable. Here we have presented a case of hemangioma occurring in a female patient in the maxillary canine-premolar region with detailed emphasis on the clinical, radiological and histopathological features.

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Hemangioma was first described in the literature by Liston (1843).
Hemangioma is the benign tumor of infancy characterized by a rapid growth phase with endothelial cell proliferation followed by gradual involution. It is reported to be occurring in 5–10% cases of 1 year old children, while few cases are reported to be congenital. The congenital forms are present at birth and may become more apparent throughout life. The peak incidence of hemangioma is in the second and fifth decades of life. Hemangiomas are more common in females than males (2:1), and more in whites than other racial groups. Though the head and neck are considered to be the most common locations accounting for 60% of cases, it is relatively rare in the oral cavity. They may be cutaneous occurring in the skin, lips and deeper structures, mucosal occurring in the mucosal lining of the oral cavity, intramuscular involving the masseter and perioral muscles and intraosseous involving the jaw bones like the maxilla and mandible. The origin of hemangioma is debatable. While some authors believe it to be a true neoplasm, others are of the view that it is a hamartoma due to its great resemblance with normal vessels and limited growth potential.

The capillary variant of intraosseous hemangioma is sparsely reported in the literature. Therefore, the aim of this paper is to present a case of central capillary hemangioma occurring in a female patient with special emphasis on the clinical, radiological and different histopathological features.

### 2. Case presentation

A female patient aged 25 years reported to the Dept. of Oral Pathology with the chief complaint of swelling in the upper gingiva which was slowly increasing in size since last 3 months. There were no relevant medical and family histories, no deleterious oral habits.

On clinical examination no significant extra-oral finding was noted. Intra-orally a diffuse erythematous growth arising from the facial aspect of the maxillary gingiva was noted in relation to 23–25. The surface of the lesion was smooth and shiny with loss of stippling of the involved gingiva. No ulceration was seen. On palpation the swelling was bony hard and non tender. No pulsation or bruit was noted. The lesion bled on provocation. There was a grade I mobility of regional teeth. (Figs. 1 and 2).

Intra-oral periapical (IOPA) X-ray revealed the presence of an ill-defined radiolucency involving the periapices of 23–25 with slight displacement of regional teeth. There was no significant bone destruction (Fig. 3).

Bone was performed under local anesthesia. The tissue was preserved in 10% formalin and sent for histopathological examination.

Sections stained with H&E revealed the presence of large number of young proliferating blood filled capillary spaces some of which were comparatively larger in size. These spaces were lined by a single layer of well formed flattened endothelial

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**Figure 1** Showing no extraoral findings.

**Figure 2** Intra-orally a bony hard swelling with a diffuse erythematous growth arising from the maxillary gingiva was noted in relation to 23–25, with no ulceration of the region and a grade I mobility of regional teeth.

**Figure 3** IOPA X-ray revealed the presence of radiolucency in the periapices of 23–25 with no significant bone destruction.
cells and lacked muscular coat. Connective tissue stroma was sparsely cellular with few inflammatory cells and no intercellular edema was noted. No nerve bundles were seen. Hence the diagnosis of capillary hemangioma was given histopathologically (Fig. 4). In proliferative phase, a large number of mast cells are a constant feature of hemangioma, hence we had performed special staining with toluidine blue which revealed the presence of mast cells (Fig. 5). In the present case, the connective tissue stoma was sparsely cellular with normal count of mast cells, hence it was concluded not to be in the stage of proliferation.

Digital subtraction angiography was performed and revealed abnormal arterial phase tumor blush or capillary hemangioma in the left upper jaw measuring about 3.3 cm × 2.4 cm × 2.3 cm in dimension. The tumor blush was fed by the sphenopalatine and descending palatine branch of the left internal maxillary artery (Fig. 6).

The patient was sent to the Dept. of oral surgery for further treatment and was asked to come after surgery for a regular follow-up.

Figure 4 Revealed the presence of large number of proliferating blood filled capillary spaces lined by flattened endothelial cells. Connective tissue stroma was sparsely cellular with few inflammatory cells.

Figure 5 Special staining with toluidine blue revealed the presence of mast cells.

3. Discussion

Hemangioma is considered to be one of the most common soft tissue tumors of the head and neck region. However, intra oral central hemangioma is somewhat rare accounting to less than 1% of all intraosseous tumors. The occurrence of central or intraosseous hemangiomas is not well documented in the literature as compared to the soft tissue counterpart. The history of hemangioma dates back to 1843 when Liston first described the case of hemangioma. Later on cases of vertebral hemangioma (Virchow, 1867), cutaneous hemangioma (Kasabach and Merrit, 1940) were documented. In 1973 Sznajder et al. described hemangioma under the term “Hemorrhagic hemangioma”.

Intraosseous hemangioma is more common in the mandible than the maxilla. The peak incidence of occurrence is the second decade of life with slight female predilection. The most common site of occurrence is body of the mandible and posterior region in case of the maxilla. Cases of condylar hemangiomas are also reported in the literature.

The origin of central or intraosseous hemangioma is still debatable. While some believe it to be hamartomatous lesion originating from the mesodermal cells that undergo endothelial differentiation, others believe it to be a true neoplasm. Malignant transformation has also been reported in some cases of central hemangiomas.

Central hemangiomas have said to present varied signs and symptoms. The patient may be completely asymptomatic or may experience mild discomfort, bleeding from the gingiva, bluish discoloration of the gingiva, displacement of teeth, derangement of the dental arch and mobile teeth. The dentist must be aware of the lesion as severe bleeding may be encountered while tooth extraction or biopsy even resulting in death of the patient. Cases of spontaneous bleeding have also been reported. In the present case, erythematous nodular growth was noted in the maxillary gingiva in relation to 23–25 which bled on provocation.
The radiological appearance of the central hemangiomas is definitely not pathognomonic. There may not be any manifestation in the radiographs in case of early lesions. In some cases there may be alteration in trabecular pattern ranging from thin or lost in some areas to thicker or coarser. Some lesions present a honeycombed appearance, sometimes with radiating spicules at the expanded periphery forming a ‘sunburst’ appearance as seen in osteosarcoma. Radiographic appearance similar to that of a giant cell lesion or an ameloblastoma has also been reported in few cases. In hemangiomas, enlargement of the involved bone is sometimes reported. The case mentioned here demonstrates the feature of enlargement of the bone.

The CT-scan which allows clear visualization of cortical involvement is useful to define the extension of the hemangioma and its relationship with surrounding soft tissues. The classical CT scan feature is the “polka-dot” appearance. Histopathologically, hemangiomas are classified as cavernous or capillary type according to their vascular network. Early hemangioma is characterized by the presence of plump endothelial cells and indistinct vascular lumen and is often known as tuveune or cellular hemangioma. Thickened multilaminated endothelial basement membrane with ready incorporation of tritiated thymidine in endothelial cells and normal count of mast cells are noted in the proliferating phase of hemangioma. As it matures, the endothelial cells become flattened and tiny capillary sized vascular spaces become prominent. When the lesion involutes, the vascular spaces become more dilated (cavernous) and widely spaced. There is little or no incorporation of tritiated thymidine in endothelial cells and normal count of mast cells are noted in the involuting phase. Capillary hemangiomas are usually present at birth, while most cavernous hemangiomas occur in adulthood. Cavernous hemangiomas are less common than capillary hemangiomas in all other areas of the body except in the oral cavity. Almost all intraosseous hemangiomas of the facial skeleton are to be cavernous type. However, the case presented by us in this paper is a type of central capillary hemangioma which is relatively rare in the oral region.

A number of growth factors including vascular endothelial growth factor VEGF, basic fibroblast growth factor [bFGF], transforming growth factor-beta [TGF-beta] and interleukin-6 [IL-6] have been described as angiogenic factors. Cellular markers like TIMP-1, bFGF, proliferating cell nuclear antigen, type IV collagenase and urokinase have also been demonstrated.

The central hemangioma of the bone may be clinically and radiographically similar to other vascular condition like central arteriovenous fistula, shunt or aneurysm. Angiography has been used as a diagnostic tool for demonstrating the presence of a vascular lesion like hemangiomas and delineating its boundaries. Also it differentiates between arteriovenous fistula and central hemangioma. Digital subtraction angiography subtracts the objects from an image that may be obstructing the area we are more interested in viewing. It reduces the procedural time. Hence in the present case, we had performed digital subtraction angiography to demonstrate the tumor blush, in the left maxillary region in relation to 23–25, fed by the branches of the left internal maxillary artery.

Hemangiomas of the jaw bones may turn out to be fatal especially in cases of close proximity to the tooth. Hemorrhage may be encountered while tooth extraction leading to death. Many congenital hemangiomas have been reported to undergo spontaneous regression at an early stage. Treatment is indicated only under some conditions where esthetic disfigurement, repetitive bleeding and palpable mass are a matter of concern. Therapeutic alternatives including surgery, radiotherapy (external radiation or radium), sclerosing agents, such as sodium morrhuate or psylliate injected into the lesion, curettage and embolization, carbon dioxide snow, cryotherapy and compression may also be tried depending upon the situation. However wide surgical excision involving the normal surrounding bone and ligation of nutrient vessels if any, will be the elective treatment.

4. Conclusion

In conclusion, it can be pointed out that though intraosseous hemangiomas account for only less than 1% of intraosseous tumors, it may be a cause of death due to hemorrhage even in minor dental procedures like tooth extraction. Here lies the importance of this case presentation where we have performed digital subtraction angiography and staining with special stains like toluidine blue which may be useful for confirming the nature of the lesion and to determine the extent and flow characteristic. This may be helpful in designing the treatment plan and preventing any fatal outcome per-operatively.

Conflict of interest

None declared.

References