

Clinical, Radiographic and Histological Assessments of a Rare Case of Intraosseous Arteriovenous Malformation in Maxilla Occurred During Pregnancy: A Case Report

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Abstract

Objective: Intraosseous Arteriovenous malformation (AVM) is a vascular hamartoma with almost 50% of cases occurring in the head and neck. These lesions are of great importance as they may cause massive bleeding during tooth extraction and surgeries.

Case report: A 33 year old female complained of swelling and gingival bleeding on the right side of the maxilla and hard palate. The swelling was noticed during pregnancy and the patient underwent a surgical excision 6 months after labor. The specimen was submitted for histological examination. An intraosseous AVM presenting in maxilla was diagnosed.

Conclusion: Although rare, intraosseous AVM may occur. The role of pregnancy and hormonal changes is still controversial. Clinical, radiographic and histological characteristics of AVM should be thoroughly assessed before surgical excision.

Keywords: Arteriovenous Malformations; Maxilla; Cone-Beam Computed Tomography; Pregnancy

Introduction

Arteriovenous malformations (AVMs) are uncommon hamartoma and almost 50% occurring in the head and neck (1). AVM is not a neoplasm, it is direct communication between artery and vein

bypassing the capillary system. They may have slow blood flow or fast blood flow. The etiology may be trauma, infection, hormonal changes or in rare instances developmental anomaly (2). AVM may form centrally or peripherally. Central lesion may cause expansion of bone, perforation and extends to the overlying soft tissues. Soft tissue discoloration to red and purple may happen (3). Central AVMs in

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the jaw may be completely asymptomatic or may cause bruit, dental loosening, gingival bleeding, cortical expansion and dysesthesia (4). The lesion has a prevalence in females with peak incidence in the second decade. Mandible is the most predominant jaw with posterior regions and retromolar area and ramus the most common sites (5). However, in rare cases, intraosseous lesion in maxilla occurs (4).

Radiographically, AVM of jaw may have various features, therefore, they are referred to as great imitators appearing as either cystic lesion or destructive malignancy. The lesion may be completely radiolucent or mixed radiolucent and radiopaque. Multi-locular, soap bubble or honey combed lesions are often seen due to the tortuous path of enlarged vessels in the bone. The borders may be well defined or ill defined. Maxillary cases may cause enlargement of the superior alveolar, infraorbital, descending palatal, and sphenopalatine arteries (6).

Cone Beam Computed Tomography (CBCT) is helpful for better detecting the location and extent of the localized jaw lesions three dimensionally, especially if surgery is needed. In addition, CBCT has significantly lower dose in comparison to Maxillofacial MDCT (7). As with hemangiomas, contrast-enhanced MDCT or MR angiography are modalities of choice to detect the feeder vessels in high flow cases.

Case report

A 33 year old female was presented in an academic center affiliated to Shahid Beheshti University of Medical Sciences, Tehran, Iran, with the chief complaint of swelling in the right maxilla and palate and bleeding from the gingiva from a year ago. The patient had a clear medical history, consumed no drugs nor medication and had a recent delivery with no complications.

On extra-oral examination, mild facial asymmetry was noticed, presenting as slight cheek swelling on the right side. Intra-oral examination showed buccal and palatal dome- shape bony hard expansion at the region of right first and second premolars (Figure 1 A). The lesion size was estimated 1.5×2 cm. The buccal mucosa was slightly red compared to the other side of the jaw. Patient complained of mild tenderness during palpation of the region. Mild gingival bleeding was seen at the cervical edge of premolars after palpation of the area, however, no

pulsation or thrill was detected. Vitality tests of the maxillary teeth on the right side showed a positive response. Conventional panoramic and intraoral periapical radiography was captured from the area (Figure 1 B and 1 C). On panoramic radiography, slight trabecular alteration at the region of right canine and premolar teeth was detected. The borders of lesion could not be easily detected. Cranio-caudal enlargement of alveolar crest at the region of right canine and first premolar was also noticeable compared to the left side. Root dilacerations between canine and premolar teeth were detected.

On periapical radiography, trabecular alteration presented as slight radiolucency with a radiopaque cortex at the periphery of the lesion at the region of premolar teeth. Root resorption was detected at the apical region of first and second premolars. For a better assessment of the extension of the lesion, CBCT scan (Newtom VGi, (Verona, Italy); FOV 8×10 , Voxel size: 0.075 mm) was requested. Reformatted panoramic, cross-sectional and axial CBCT images show buccal and palatal expansion (Figure 1 D). Thinning and perforation of the cortex were suggested on buccal cortex. Slight spiculated periosteal reactions were detected on the palatal surface.

Based on the patient history of gingival bleeding, and clinical and radiologic examination, a provisional diagnosis of vascular lesion was given. Needle aspiration presented low flow of blood.

The patient was further referred to the department of oral and maxillofacial surgery and the lesion was excised surgically under general anesthesia in the operating room. The lesion was approached with a sulcular incision from canine to second molar (Figure 1 E). The surgical view revealed a spherical red pulsatile lesion resembling a typical vascular lesion approximately $3 \text{ cm} \times 2 \text{ cm} \times 0.5 \text{ cm}$. Due to the controlled nature of bleeding, no need for catheterization and embolization were required. The histopathologic Hematoxylin and Eosin (H & E) examination confirmed numerous dilated blood vessels and unremarkable bony trabecula (Figure 1 F). Based on clinical, radiographic and histopathologic findings and according to The International Society for the Study of Vascular Anomalies (ISSVA) 2018 classification, the diagnosis is capillary-venous malformation (CVM). The patient experienced immediate improvement, and postoperative one and three month follow up of patient was uneventful. The area had healed with no complication.

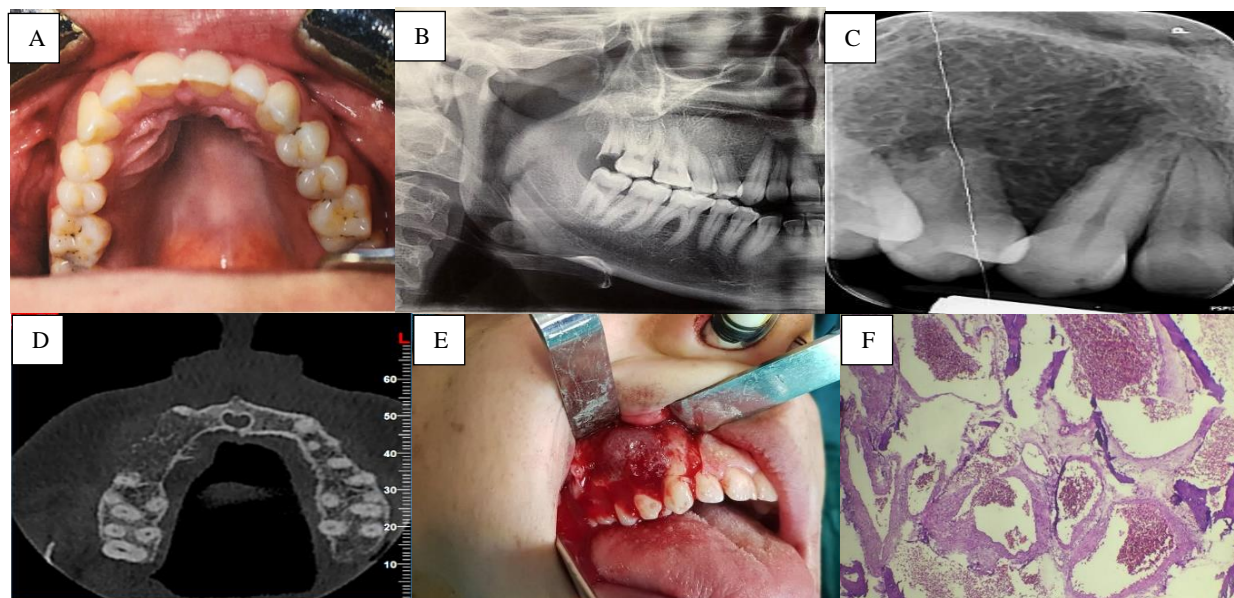


Figure-1: (A) Intra-oral occlusal view of the lesion show spherical expansion of buccal and palatal cortex at the site of right canine and premolars. (B) Cropped panoramic radiography shows altered trabecular bone pattern at the region of upper right canine and premolars. Note the dilaceration between teeth roots and slight distal rotation of premolars. (C) Intra-oral periapical radiography of maxillary right premolars show a mixed radiolucent radiopaque lesion with corticated border at the periphery. Note the loss of lamina dura at the apices of premolars and canine and root resorption at the apice of first premolar. (D) Axial Cone Beam Computed Tomography images show thinning and perforation of buccal cortex. Note the presence of spiculated bone reaction on palatal cortex which may happen in vascular lesions. (E) Intraoperative appearance of the mass being exposed. (F) Histological features of intraosseous arteriovenous malformation of maxilla show vascular spaces of variable sizes filled with RBCs and lined by endothelial cells. The spaces are separated by fibrous connective tissue and bony trabeculae (H & E stain x40)

Discussion

Arteriovenous malformation may occur in any part of the body but more than 50% are found in head and neck. These lesions often occur in the soft tissue and are rarely seen intraosseous and are differentiated based on clinical manifestations and histopathologically according to IVSSA 2018 classification (8). Although AVM occurs more commonly in the mandible than maxilla by a factor of 2 to 1 (9), in this report, the lesion was located in the maxilla. Similarly, in the study of Katcher et al., arterial malformation in the posterior maxilla of an 8 year old boy was detected (10).

The most common signs and symptoms of AVM in jaws are soft tissue swelling, pain, paresthesia, facial asymmetry, mobility and displacement of teeth. The patient may complain from gingival oozing and bleeding, and fell pulsation. The overlying mucosa is usually discolored to reddish blue or purple (1, 11, 12).

On conventional radiographies such as panoramic view, bone resorption or bone formation may both be seen therefore the lesion varies from radiolucent to mix radiolucent and radiopaque. Computed tomography and cross sectional imaging present accurate trabecular bone patterns (13) as in this case, CBCT imaging could accurately show the extent of the lesion and its effects on the surrounding structure. AVM may cause expansion of neural and artery canals. This expansion may be in a tortuous or serpiginous pattern. Neural lesions such as Schwannoma or Neurofibroma may also cause expansion of these canals, however, the expansion is more uniform and fusiform in shape (14).

The most common clinical and radiographic differential diagnosis of AVM, is hemangiomas (1), Hemangiomas are benign vascular tumors in infancy that have rapid growth followed by gradual involution. Histopathologically, hemangioma is a benign tumor, in early phase composed of plump

endothelial cells and often indistinct vascular lumen. Vascular malformations don't show active endothelial cell proliferation and vascular channels resemble the origin vessels. Due to the number of radiographic appearances, cystic and multilocular lesions may also be included. When an intraosseous AVM extends beyond the bone surface and displaces the periosteum, linear spicules of bone can be seen emanating from bone in a sunray-like pattern which is commonly seen in malignant osseous lesions such as osteosarcoma (3, 14).

The preferred imaging is contrast-enhanced MDCT, or conventional and MR angiography, especially in high flow lesions, to better assess the feeder vessels and treatment options (15-17).

In this report, the patient was recently pregnant, noticing the lesion during and after pregnancy, and previous studies suggest that pregnancy can lead to development and progression of AVMs (12, 16). The estrogen and progesterone hormonal changes during pregnancy added to the changes in blood flow maybe the probable cause, although this is not definite. Many AVMs tend to go under postpartum regression (11). Martines et al indicated that pregnancy can have marked adverse effects on vascular malformations. They reported an unusual case of an AVM of the base of tongue in a 32-year-old primigravida at 23.2 weeks of gestation (18).

The treatment for large high flow AVM, requires multidisciplinary team approach. Arterial embolization, interventional management and surgical treatment are different treatment modalities (17). Radiotherapy, sclerotherapy, curettage and dehydrated alcohol are other treatment modalities and may decrease blood flow in selected cases(15, 19). In this case, the lesion was small, low flow with no prominent feeder vessels and therefore ligation and embolization were not necessary.

Conclusion

Although rarely encountered, AVMs should be carefully examined clinically and through imaging, before any treatment modality is applied to avoid bleeding and life threatening complications.

Conflict of Interests

Authors have no conflict of interests.

Acknowledgments

The authors declare that there is no conflict of interest regarding the publication of this article.

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