REVIEW ARTICLE

Arteriovenous Malformations of The Mandible and The Maxilla: A Review

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ABSTRACT

Arteriovenous malformations (AVMs) are uncommon vascular lesions occurring anywhere in the body and can be life threatening due to potential massive haemorrhage. Vascular malformations of the jaws can lead to disastrous complications. The literature presents the pathophysiology and clinical aspects of these lesions, as well as the divergent views of the authors. Traditional treatment involved extensive surgical resection of the mandible. Catheterization and embolization, with direct trans osseous injection of cyanoacrylate, appears to be the least harmful treatment in certain conditions. Endovascular techniques, interventional radiology are now the best method to control active hemorrhage and ultimately cure these lesions.

Keywords: Arteriovenous malformations, Embolization, Jaws, abnormalities.


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INTRODUCTION

Before the 1980s, vascular lesions were referred to as “haemangiomas.”¹ Thereafter, they were subdivided into hemangiomas and vascular malformations.² According to Mulliken and Young, two types of vascular lesions can be recognized, which depend on the intrinsic properties of endovascular cells, namely haemangiomas and vascular malformations.³ Haemangiomas demonstrate endothelial hyperplasia and enlarge by cellular proliferation. Clinically, haemangiomas usually appear in early infancy, grow rapidly during the first months of life, then slowly involuting over 5 or 6 years.⁴,⁵ While vascular malformations enlarge with the growth of the child and do not undergo spontaneous involution. Vascular malformations are subdivided based on blood flow rate: “slow flow” (capillary, venous, lymphatic or mixed) versus “fast flow” (arteriole, arteriovenous, fistulae or shunt) subtypes.⁶ AVM are errors that are present at birth and become prominent due to an event like surgery, trauma and infection. Most common signs and symptoms of these patients, in the mixed dentition period, is spontaneous hemorrhage along with hyper mobility of the teeth from the surrounding gingival sulcus.⁷ Although vascular malformation is rare, the dentist must always able to recognize the clinical sign and symptoms for propose proper treatment. Various terms are used to refer the lesion, notably, cavernous hemangioma, arteriovenous aneurysm, pulsatile hemangioma, angioma, arteriovenous shunt, central hemangioma, arteriovenous malformation vascular malformation and arteriovenous fistula.¹ Colour Doppler ultrasonography usually helps in differentiating between these subtypes. By which one can determine flow rates.⁷ Other imaging modalities used for diagnosing vascular malformations include MRI which helps in to evaluate the relation of the lesion to the surrounding tissues. The other major diagnostic tool like Magnetic resonance angiography provides detailed information regarding flow characteristics and the extent of local tissue involvement.⁸ Plain radiographs are used for the detection of vascular malformations; which showed cortical erosion of bone in approximately 6% of cases or calcified phleboliths.⁹ The management of vascular lesions depends upon the lesion’s location, blood flow characteristics, symptoms, functional disability and cosmetic deformity.¹⁰ Treatment may be surgical or non-surgical. The latter includes intravascular embolization with coil and/or sclerosing solutions. Surgical resection is reserved for lesions that are extensive and/or refractory to endovascular therapy. Cure is defined as the complete eradication of disease or permanent resolution of symptoms with complete devascularization.¹¹,¹² Sclerosing agents cause a thrombosis or marked tissue irritation with local inflammation and tissue necrosis resulting in tissue contraction and fibrosis. Some agents include sodium morrhuate, nitrogen mustard, boiling water and sodium tet-
radecyl sulphate. They have been used both to treat and for embolization of high flow vascular malformation and symptomatic hemangiomas.13,16

Etiopathogenesis
The blood shunted to the malformation causes the lesion to grow, which in turn causes increased shunting of the blood; hence, a vicious circle. Blakey G and Kula K, states that lesions may occur due to an abnormality of angiogenesis of the vascular system and Hemangiomas may be caused by a failure of differentiation in the early stages of angiogenesis.1 The appearance of lesions are more commonly in childhood and tend to regress or disappear in adolescence which are rarely associated with fatal hemorrhages.15 VMs, are caused by a disturbance in the late stages of angiogenesis and result in the persistence of arteriovenous anastomoses present during embryonic life.16 They may be lymphatic, capillary, arterial, venous, or mixed. Arteriovenous origin are often known as “high-flow vascular malformations” and are often the cause of massive, sometimes fatal hemorrhages.15 VMs, which usually present as developmental anomalies from birth, develop in proportion to physical growth.17 Increment in size the of these vascular malformations, are imperceptible at an early age, which is promoted by local hemodynamic factors. Low vascular resistance usually causes a shunting of the blood which decreased perfusion of the peripheral tissue, flow, gradual dilatation of the nutrient arteries with atrophy of their musculo-elastic wall and decreased resistance, and dilatation and arterialization of the draining veins, owing to the increase in intraluminal pressure.16 The blood shunted to the malformation causes the lesion to grow, which in turn causes increased shunting of the blood; hence, a vicious circle.

Clinical signs
Vascular malformations are frequently seen in the skin, but rarely affect the visceral organs or bones; approximately 51% occur in the head and neck, with a male female ratio of 1 to 1.5,18,19 Some authors have noted predominance in women (2:1), while others have reported equal prevalence among men and women.17 unlike hemangiomas, which involute, the size of AVMs generally increases in size proportionately as the child grows. The mean age at presentation is 19 years with equal predilection for both males and females.20 Jackson IT, Jack CR et al state that AVMs in the maxillofacial skeleton are common with approximately 31% presenting in the head and neck20 while Persky MS concluded that About 50% of all bone involvement occur in the skull and the maxillo-facial area.18

Mandibular VMs usually appear during adolescence, with extremes at 3 months and 74 years of age.16 Lesions of the mandible are rare and potentially life threatening entities that can present as innocuous episodes of gingival bleeding, slow-growing expansile masses, or severe haemorrhage. A biopsy or even a simple tooth extraction can cause a catastrophic bleeding that may even lead to death.15-21 Gingival bleeding are the symptom which are more and mostly documented cases.22 Massive hemorrhage, even exsanguination, have been documented following the extraction of teeth associated with these VMs.15,22 Central lesions are painful and produces an alteration in facial morphology.17 Vascular naevi or phlebectasias may causes the dis-coloration of the adjacent mucosa or skin.16,22 At the level of the nose there may be nasal blockage, epistaxis, rhinitis, sinusitis, proptosis or diplopia.17 Cardiac symptoms are rare.16,22

Summary of clinical signs
1. AVM of the jaws is uncommon. Mandible - more (twice than Maxilla), Maxilla - less.
2. Pain
3. Erythematous gingiva.
4. Spontaneous gingival bleeding.
5. Resorption and mobility of teeth.
6. Soft tissue discoloration, facial swelling, and asymmetry.
7. Bruits or pulsations can be detected in large lesions.
8. The most severe hazard with AVM lesions is potentially profuse and uncontrollable bleeding when performing dental procedures such as tooth extraction, and biopsy or incision of a suspected cyst and with primary tooth exfoliation.

Radiological signs
In the maxilla and the mandible, lesions are produced by a poorly defined, radiolucent image.17 The appearance of a honeycomb or soap bubbles, with small rounded and irregular lacunae.16,22,23 Magnetic resonance and CT scanning helps in to clarify the extent of the lesion, bone erosion and the involvement of major vessels.17 Super-selective arteriography is an essential tools for the identification of the VM and contributory vessels.24 In this technique the radiopaque substance is injecting into the vascular system through a catheter near the region. After that image is processed by the computer, and the bone densities are subtracted for a clearer illustration of the vascular system. Arteriography of the external carotid must be done bilaterally, which gives the importance of the collaterals and multiple anastomoses of the maxillary artery.24

Treatment
Arterial embolization is the treatment of choice and can be repeated in the case of relapse. Intralesional injection of sclerosing agents can be used in the attempt to reduce the number of arterial embolizations. In AVMs of the mandible, surgery should be reserved only for cases that are refractory to endovascular therapy and/or for therapeutic complications that are not otherwise treatable (bone fractures and/or necrosis). In AVMs of the soft tissue, surgery could be used if there...
are no significant side effects. Lastly, the interval between embolization and surgery should be as short as possible, and MRI and MRA should be repeated before surgery.

Divergent views of the authors
In a presented case a 6 year old patient with mandibular arteriovenous malformation who had hemorrhagic shock after a tooth extraction. The patient gets treated with transarterial and direct intralesional embolization but local infection occurred 3 months after embolization, which soon controlled. During follow up examination Serial computed tomography and panoramic radiographs were performed. The imaging studies showed gradual obliteration of the vascular space followed by normal bone regeneration and remodeling. All the processes were completed during a period of two years. There was no signs of recurrence of the vascular malformation and no any abnormal growth was seen in the right side of the mandible after four years and four months of observations.

Lt Col Suresh Menon, Maj SK Roy Chowdhury, Col Chandrak Mohan, in their study reported a 14 year old boy having complained of mobility of Mandibular left 1st and 2nd molars since a couple of months. Clinical examination showed a good periodontal condition with mobility of the teeth in question. Ortho Pan Tomograph showed a diffuse radiolucency of the left mandible with clear cut margins anteriorly in relation to the canine but with ill-defined border posteriorly in the retro molar region. A CT scan of the area showed confirmed osteolytic lesion with buccal plate expansion and having lingual plate thinning. In clinical finding there was an unusually prominent pulsation seen and bruit over the left facial artery giving rise to a doubt, whether this was a vascular lesion. So, a trans -femoral angiography was performed and confirmed the diagnosis of high flow AV malformation involving the left facial, lingual and maxillary artery. By the help of MRI it seen that there was also soft tissue involvement of the lesion and confirm the limitation of the lesion within mandible. By the help of gel foam embolisation of the facial, lingual and maxillary arteries were achieved and poly vinyl alchohol and the occlusion confirmed by post embolization angiography. After two days the resection of the lesion was done but a preoperative angiography revealed recanalization of the vessels filling the lesion. Again the embolisation was done and the patient taken up for resection the next day. After that Hemimandibulectomy was performed and an immediate reconstruction using iliac graft was done. Gaétan Noreau, Pierre-É. Landry and Dany Morais conducted a study in a 9-year-old patient for assessment of an asymptomatic left mandibular intraosseous lesion. Slight extrusion and dental mobility with a non-hemorrhagic, gingival granulomatous swelling without vascular bruit was detected. The panoramic examination showed radioluency of tooth 34 extending to the region of the lingula and the neck of the condyle and root resorption was also noted in tooth 36. The CT scan confirmed erosion at the lingual cortex. The simple extraction of tooth 36 caused a massive hemorrhage. The patient was discharged, but was readmitted in emergency for treatment of massive and spontaneous gingival hemorrhage. A cerebral and facial angiography, revealed the arteriovenous malformation of the left mandibular body. Embolization was done by direct puncture of the lingual mandibular cortex and injection of a mixture of cyanoacrylate and Lipiodol, under constant digital pressure at the site of the hemorrhage. Over the 6 months following the embolization Dehiscence of the mucosa, exposing the embolization material at the level of the alveolus of tooth 36, was noted. Radiographic examination showed changes suggesting a revascularization of the lesion. Bacteriological cultures confirmed major infection
with actinomyces odontolyticus. The positive gallium scintigraphy called for 4 months of oral antibiotic therapy (clindamycin). Examination at 14 months confirmed excellent gingival healing, no signs of infection, and the maintenance of facial symmetry and complete ossification of the lesion site.

CONCLUSION

There was always a high degree of suspicion leads to their diagnosis and considerably reduces the risks of a catastrophe once identified. Treatment by embolization and catheterization with intra lesion injection of cyanoacrylate which allows for conservative anatomic and functional recovery. This technique is relatively non-invasive and safe.

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