

Arteriovenous Malformation Of Mandible With A Diverse Radiographic Appearance-A Case Report

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Abstract:

Arteriovenous (AV) malformation is a congenital vascular anomaly in which there is an abnormal connection between the arterial and venous systems due to developmental arrest during embryogenesis. AV malformation of the head and neck are rare congenital vascular anomaly but when present, they are persistent, progressive, fatal, leading to considerable blood loss, and often incomplete resection leads to recurrence of the lesion. The current case shows AVM in a 12-year-old girl with no distinctive signs except for swelling for the past one month suggesting AVM in Right body and ramus of Mandible. Malformation was diagnosed with the help of detailed patient history, clinical evaluation & radiographs. The definitive diagnosis was made by CT angiography.

Keywords: Arteriovenous malformation, mandible, tennis racquet appearance, Computed tomographic angiography.

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I. Introduction:

Early to 1980s all vascular lesions were referred to as hemangiomas^[1]. Later in 1982, Mullikin and Glowacki classified vascular lesions into hemangioma and vascular malformations based on endothelial characteristics. AV are congenital in nature formed by mutation early in embryogenesis and later developed in life by trauma or by other endocrine dysfunction^[2]. As AVM is a benign lesion, the most common sites in the head and neck are scalp, auricle, cheek, nose, lip and mandible^[3]. Their clinical presentation may vary from onset of minor gingival bleeding, lower lip numbness, facial deformities, and even hemorrhagic shock after extraction. AVMs frequently occur without syndromic, a classical example which is related with syndrome is hereditary hemorrhagic telangiectasia^[4]. They may obtain feeders and drainers with the involvement of surrounding tissues. Here we report a case of 12 year old with AVM in mandible with an unusual radiographic appearance.

II. Case Report:

A 12 year old female child, came to our hospital with the chief complaint of swelling in right side of the face for the past one month. Past medical and family history were non contributory.

III. Clinical Findings:

On extra oral examination a facial asymmetry was present due to the swelling. On inspection, a single, ovoid, well defined swelling of size 4x3 cm size present on the right side of mandible extending antero-posteriorly 1 cm from R corner of mouth to R posterior border of ramus, superiorly from R zygomatic region inferiorly extending upto R inferior border of mandible.

On palpation, the swelling was warm, bony hard in consistency and non tender, with palpable submandibular lymph nodes which is mobile and non tender. On intra-oral examination, there was no evidence of swelling, occlusion was normal.



Caption

IV. Diagnostic Assessment:

Fig-1: Orthopantomogram revealed a multi-locular radiolucent lesion with radiopaque straight septa seen from R body to R coronoid process of mandible resembling tennis racquet appearance, suggestive of odontogenic myxoma.



Caption

On aspiration blood / blood tinged fluid with delayed clotting was obtained suggestive of vascular malformation.



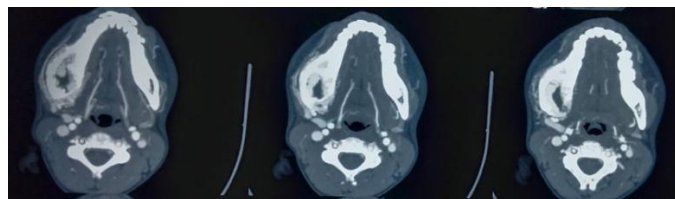
Caption

Fig 2 : on aspiration blood obtained.

Radiographically, multi-locular lesions viz. Ameloblastoma, Peripheral Giant cell granuloma, Central hemangioma were considered in the differential diagnosis.

CT-Angiogram was advised suspecting some vascular malformation.

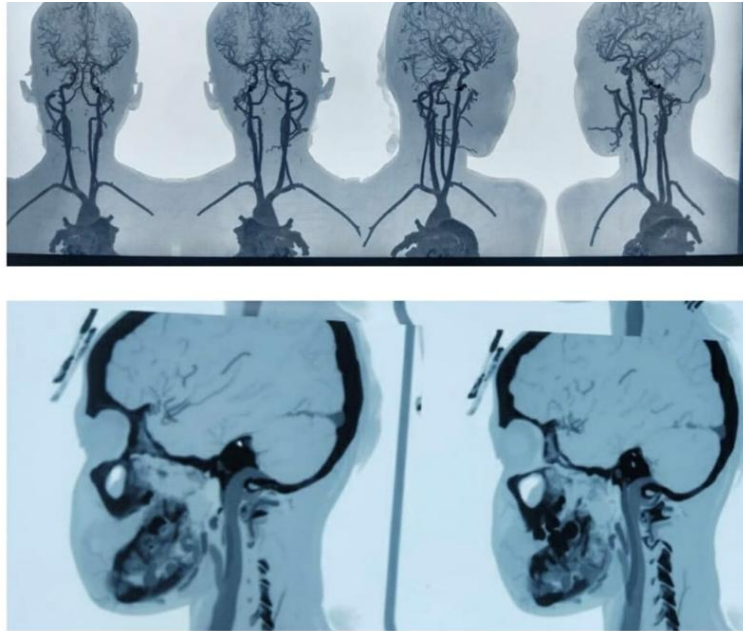
CT angiogram revealed bicortical expansion with radiopaque density seen in the R body extending to R coronoid process of mandible.



Caption

DSA cerebral angiogram was advised for further details of the course of blood vessels.

Arterial access was achieved by puncturing right femoral artery, in right groin region. Selective angiography was performed, selectively cannulating both external carotid artery (ECA) and right facial arteries.



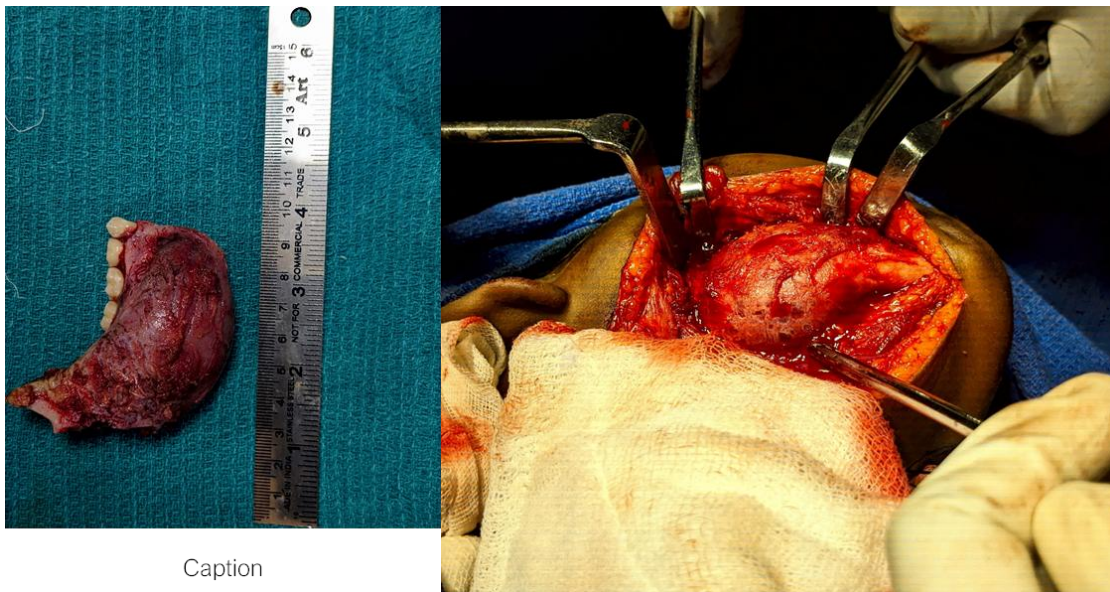
Caption

Fig-3: Abnormal hypervascular blush noted in the body of right hemi-mandible measuring 7.3x5.5x3.3cm. vascular malformation is predominately supplied by multiple feeders from right facial, right maxillary artery and lingual arteries. predominant draining vein noted with venous ectasis draining into right IJV.

Findings of angiography were suggestive of vascular malformation in right hemisphere-mandible as described. Surgical correction with ligation of major vessels was planned.

Surgical Intervention:

Patient underwent endo vascular embolisation with block resection and reconstruction with titanium plates.



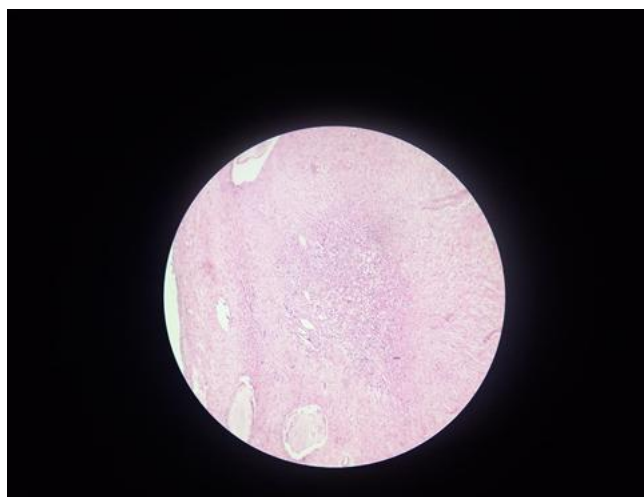
Caption

Post Operative Radiograph:



H/P REPORT:

Section showed blood vessels of varying calibers filled with RBC in a fibromyxoid connective tissue. Numerous endothelium lined small vascular spaces filled with RBC's along with endothelial cell proliferation are noted which correlated with the clinical diagnosis of Arteriovenous malformation.



Caption

Follow Up And Outcome:

Patient is still under follow up and there were no signs of recurrence after a follow up period of two years.

V. Discussion:

AVM lesions often occur in the soft tissues and are rarely observed intra-osseous and are differentiated based on clinical manifestations and histopathologically according to IVSSA 2018 classification^[5]. The detection rate of AV malformation in general population based on prospective data from New York Islands study is approximately 1.34/100,000 persons^[6]. A firm, painless, bony expansion is generally seen which can cause gross facial asymmetry and distortion of local anatomy, however pain and paresthesia are not directly linked to AVMs but can occur due to expansion and insult of adjacent structures. If it occurs in the proximity to the teeth, AVMs can cause mobility and displacement. In this present case, extra oral swelling and facial symmetry were observed without any pain / paresthesia, mobility and displacement^[7]. However, there have been reports of root resorption occurring , which can confuse the diagnosis as it is often associated with more destructive processes such as ameloblastomas or osteosarcomas. The radiographic appearance often mimics other multi-locular lesions. Generally it shows honeycomb or soap bubble appearance. It may also present sun ray appearance. However tennis racquet appearance is rare. The radiographic differential diagnosis of these lesions includes ameloblastoma, ameloblastic fibroma, odontogenic myxoma, central giant cell granuloma and metastatic malignant tumors

(Mohammadi et al., 1997)^[11]. Before performing a biopsy or surgery in a radiographically suspected case of ameloblastoma or aneurysmal bone cyst, clinician should advise a contrast CT or MRI to rule out the possibility of an AVM to avoid sudden massive haemorrhage^[8]. Contrast-enhanced CT can be useful in assessing the AVMs. Angiography is currently the gold standard diagnostic aid for determination of location and flow characteristics of vascular lesions. Angiography is useful to determine blood vessels supplying blood to the lesion, the relative venous outflow characteristics and the presence or absence of arteriovenous shunts^[9]. Definitive diagnosis can only reliably be achieved through biopsy and histopathological examination. The Histopathological features are fairly consistent with AVM histology, proliferative epithelial cells and fibrous tissue are often seen surrounding the lesion and the remaining bony trabeculae are interspersed with vascular structures and endothelial cells^[10].

VI. Conclusion:

A radiograph showing multi-locular radiolucency resembling tennis racquet appearance with radiopaque internal straight septa in jaws is usually suggestive of odontogenic myxoma, but on aspiration blood was obtained in this case, which upon further investigation with CT angiography confirmed the diagnosis of AVM of mandible. The clinician should be aware before performing a biopsy, which may lead to torrential hemorrhage and even death of the patient. A preliminary CT angiography has the advantage of providing bony details along with status of feeder vessels of the lesion which is a prerequisite for surgery or endovascular intervention. Therefore, angiography remains the diagnostic gold standard in the treatment of AVM.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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