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RESEARCH ARTICLE

ARTERIOVENOUS MALFORMATION OF THE MANDIBLE WITH CONGENITAL CARDIAC DISEASE: A RARE CASE REPORT

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ABSTRACT

Arteriovenous malformation (AVM) of the mandible is a rare and potentially life-threatening condition which can lead to massive hemorrhage. The following is a description where a large mandibular AVM presented along with the congenital cardiac disease. An orthopantomogram (OPG) was performed which was suggestive of hemangioma. A computed tomography (CT) angiography revealed a large mandibular AV malformation. It is important for both clinicians and radiologist to be aware of this type of lesion that can have life-threatening complications. It is important to define the anatomical location and the feeder vessels of the entity in detail preoperatively. This communication highlights the common differential and use of multi-detector CT angiography along with other imaging modalities to prevent a fatal hemorrhage and arrive at a correct diagnosis.

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INTRODUCTION

Arteriovenous malformations are abnormal, direct communications between arteries and veins that bypass the capillary bed. They are uncommon lesions in the head and neck. Most lesions in the jaws occur in the ramus and posterior body of the mandible. It is important to recognize the hemorrhagic potential of these lesions because extraction of a tooth adjacent to an arteriovenous malformation may result in lethal exsanguinations (Robert *et al*., 1999).

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Before the 1980s, vascular lesions were referred to as hemangioma. Mullikinand Glowacki, in 1982, classified vascular lesions into hemangioma and vascular malformation based on endothelial characteristics. Vascular malformations can be categorized into low-flow lesions (capillary malformations, lymphatic malformations andVenous malformations) and high-flow lesions [arteriovenous malformations (AVMs) and arteriovenous fistulae], according to blood flow characteristics (Mulliken, 1982). Arteriovenous malformations (AVMs) occur as a result of errors in vascular morphogenesis present at birth. These grow proportionately with age (Waner, 1995; Higuera*et al*., 2006) and manifest at any time during life due to an event like trauma, surgery,

infection, etc (Fathi *et al*., 1997). Mandibular AVMs are uncommon and potentially life-threatening vascular malformations (Anderson *et al*., 1981; Jackson, 1990). Young female patients are predominantly affected. They are frequently high-flow vascular malformations (Larsen, 1993). Clinically, they may present with minor gingival bleeding, dental loosening, lower lip numbness, facial deformity, malocclusion and sometimes hemorrhagic shock following extraction of teeth (Rodesch, 1988; Mohammadi *et al*., 1997).

Case Report

A 17 year old male patient was referred by a private practitioner to the department of oral medicine and radiology with a chief complaint of swelling on the left side of the mandible since 8 months. History revealed that swelling was noticed after trauma due to hit by a ball on the same region and increase in the size of the swelling was noticed. History of bleeding from the swelling every morning till 1week after trauma, there was no pain associated with it. The patient had given a vague medical history of cardiac disease since birth. Family history was non-contributory. Physical examination revealed the clubbing of fingers and systemic examination was remarkable with abnormality in cardiovascular system for which patient was advised for cardiac investigations. On extra oral examination facial asymmetry was noted with diffuse swelling extending antero-posteriorly from parasymphysisupto anterior border of ramus of mandible and supero-inferiorly from angle of mouth to the lower border of mandible (Fig 1). On inspection surface texture of swelling was normal with no pulsations. On palpation the temperature was raised, swelling was non- compressible, non-tender and bony hard in consistency. On intra oral examination submucosal swelling and vestibular obliteration was seen with normal occlusion (Fig 2).

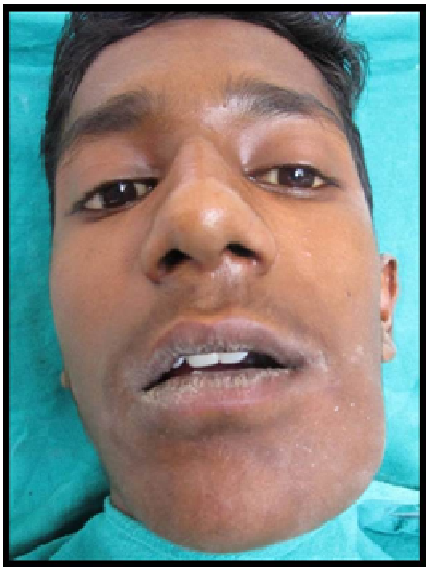


Fig-1 Swelling seen on the left side of mandible

A provisional diagnosis of hemangioma was given with differential diagnosis of AV malformation, myxoma, ameloblastoma, central giant cell granuloma. Patient was advised for radiological investigations of OPG, IOPA, lateral oblique, occlusal topography, CT scan and angiography. Panoramic radiograph showed abnormal trabeculae pattern in the region extending from the distal of tooth 33 upto distal of tooth 36 with tennis racket and honeycomb type of appearance.

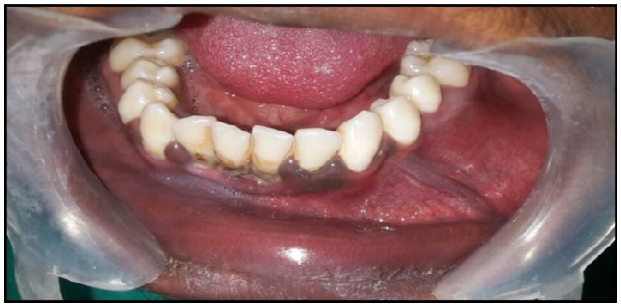


Fig. 2. Intraoral vestibular obliteration on left side

Dilaceration of the root (apical two third) of tooth 35 was seen. Extension of trabeculationsupto 1mm below the lower border of mandible was also appreciated (Fig 3). IOPA revealed curved and irregular trabeculae pattern in the region of tooth 33-36(Fig 4). Lateral oblique showed abnormal trabeculae pattern with criss-cross type of appearance in the same region (Fig 5).

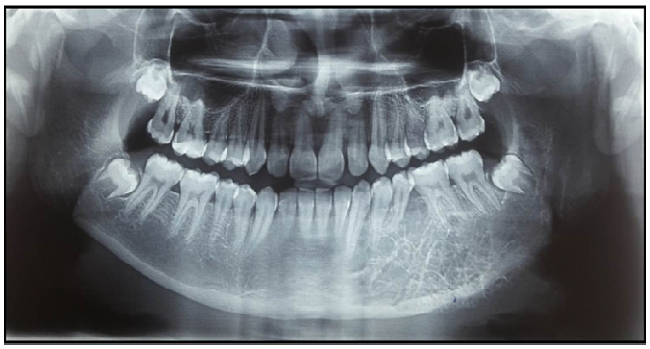
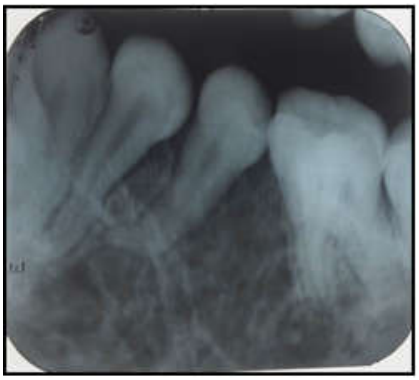
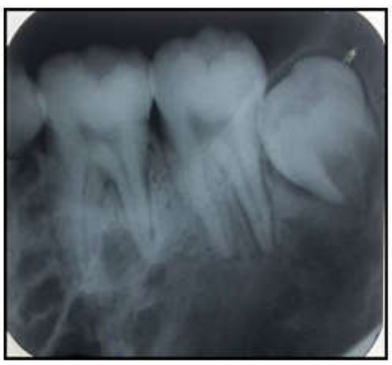


Fig-3 Panoramic radiograph shows abnormal trabeculations on the left side of mandible with extension of trabeculations below the lower border of mandible



(a)



(b)

Fig-4 (A and B) IOPAR shows curved and irregular trabeculaes in the region of 33-36

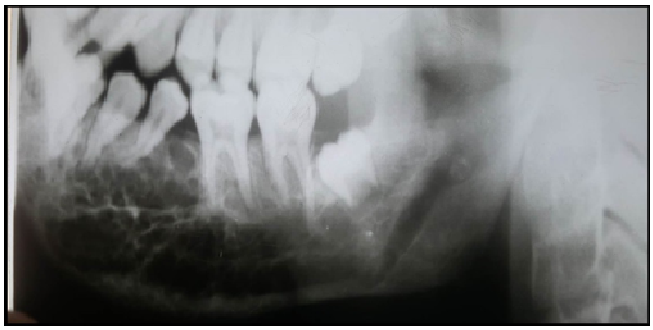


Fig. 5. Lateral oblique shows abnormal trabeculae pattern with criss-cross type of appearance on the left side of mandible

Occlusal topography revealed the expansion of buccal cortical plate with extension of trabeculationsupto 2-3mm below (Fig 6). CTscan revealed evidence of well-defined expansile lytic lesion with multilocular honeycomb like appearance in the left side of the body of mandible extending lateral to the parasymphyseal region. No calcification was seen within the mass. The cortex of lesion was continuous with the cortex of the mandible. The lesion measured 2.8-4.4cm. The cortical bone was expanded, eroded and showed breach at places on the medial and extensively on the lateral walls (Fig 7). CT angiography was done in arterial phase. Lesion showed intense arterial supply from branches of left external carotid artery mainly facial artery with appearance of early draining veins consistent with tumoral AV shunting. The facial artery was engorged with tortuous branches.

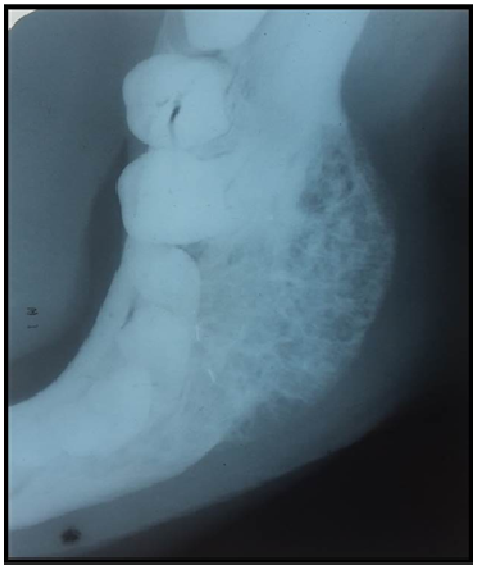
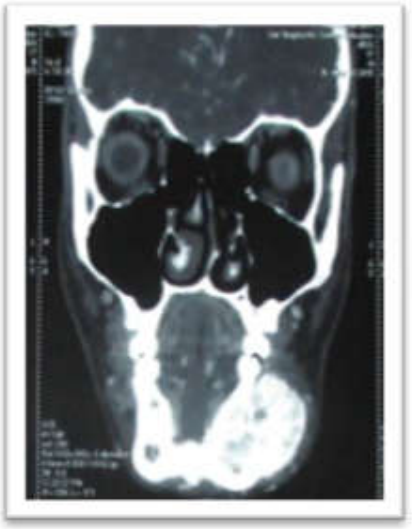
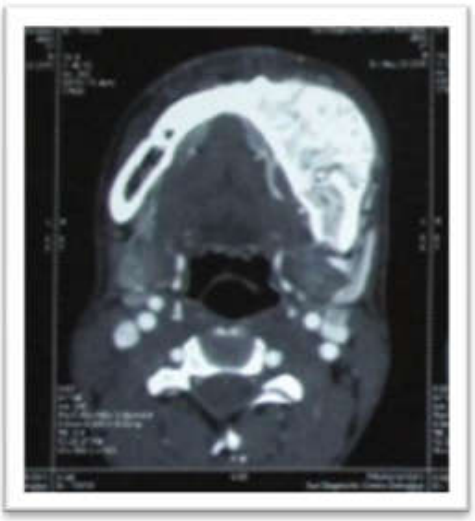


Fig. 6. Occlusal topography revealed the expansion of buccal cortical plate with extension of trabeculationsupto 2-3mm below

Early draining veins were seen. Image morphology suggested of an aggressive vascular lytic bony lesion. Biopsy correlation was suggested (Fig 8). All radiological investigations confirmed the diagnosis for AV malformation of the left mandibular region. Patient was further advised for cardiac investigations of chest x-ray, ECG and cardiac ultrasonography as he had given the vague history of cardiac disease and moreover cardiovascular abnormality was detected on examination with the clubbing of fingers. Chest x ray revealed mild cardiomegaly. ECG was abnormal with sinus bradycardia, right arterial enlargement, right axis deviation, right ventricular hypertrophy.



(a)



(b)

Fig-7 (A and B) CTscan revealed evidence of well-defined expansile lytic lesion with multilocular honeycomb like appearance in the left side of the body of mandible

Cardiac ultrasonography revealed aortic enlargement, aortic septal discontinuity, right ventricular outflow tract was narrowed, main pulmonary and its branches were hypo plastic. Suggested of Tetralogy Of fallots.

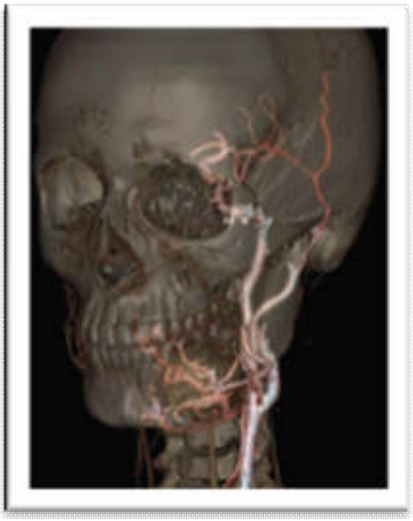
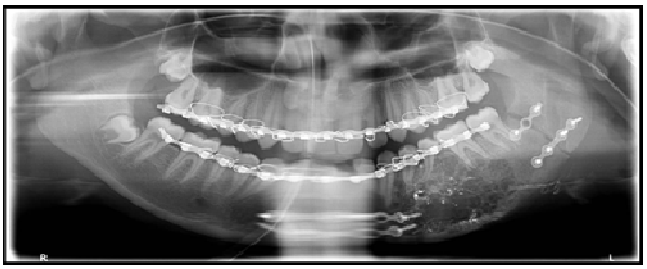


Fig. 8. CT angiography shows aggressive vascular lytic bony lesion alongwith intense arterial supply from branches of left external carotid artery mainly facial artery

(a)



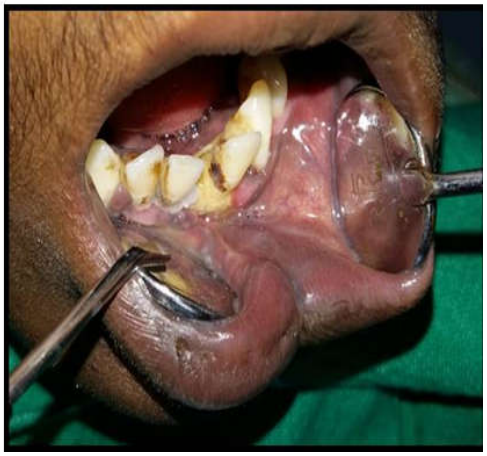
(b)

Fig-9 (A and B). Post-operative photographs after the complete excision of the lesion alongwith the arch bar wire fixation of the mandible

All cardiac investigations confirmed the diagnosis for congenital cardiac disease. So the final diagnosis of AV malformation of the left mandibular region with congenital cardiac disease was given. Treatment included complete surgical excision of the lesion after the ligation of facial artery as it was the feeder artery in angiography after which the mandible was sectioned in midline and at angle region. Bony window was created, which was filled with hydroxyapatite granules later. The mandible was then fixed with 3 and 4 hole plates at symphysis region, 3 and 4 hole plates at angle of mandible (Fig 9). Arch bar wire fixation was done which was released after a period of 1month. Postoperative recovery was uneventful with complete resolution of the swelling along with residual facial scar (Fig 10), however the patient was unconcerned and declined any future surgery. A 3month postoperative review revealed no recurrence. The excised sample was sent for histopathological examination which confirmed the diagnosis of AVmalformation as one of the fragment showed varying sized arteries and veins, feeding artery was also identified at the periphery showing the thrombus. Overall features were consistent with arteriovenous malformation.

DISCUSSION

Approximately 51% of vascular malformations occur in the head and neck region and the male-to-female ratio is 1:1.5 (Nekooei *et al*., 2006). Extra cranial AVMs of the head and neck are high-flow lesions and among the most serious of the vascular malformations because they are difficult to diagnose, treat and cure (Stapf *et al*., 2003). Multiple imaging modalities should be used to evaluate characteristics of AVMs such as size, flow velocity, flow direction, relation to the surrounding structures and lesional contents (Hyodoh *et al*., 2005).



(a)

(b)

Fig-10(A and B). Postoperative recovery with complete resolution of the swelling along with residual facial scar

There are no pathognomonic radiographic features to distinguish AVMs on plain radiographs. They may appear as bone erosions, sclerotic changes, periosteal reactions or cyst-like radiolucent lesions. A sunburst effect, created by spicules radiating from the center, is often present (Hyodoh *et al*., 2005). The radiographic differential diagnosis of these lesions includes ameloblastoma, ameloblastic fibroma, odontogenicmyxoma, central giant cell granuloma and metastatic malignant tumors (Mohammadi *et al*., 1997). 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 hemorrhage from the lesion and mortality and morbidity associated with it. Contrast-enhanced CT can be useful in assessing the AVMs. The drawbacks of CT are considerable exposure to ionizing radiation and limited information about blood flow.[13]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 (Nekooei, 2016). Super selective arteriography remains an essential tool in the identification of an AVM and contributory vessels.[14]Super selective angiography is an invasive procedure and not available everywhere.

CT scanning and MRI are sufficient in most cases to clarify the extent of the lesion, bone erosion and involvement of major vessels, feeder artery and draining vein (Johnson, 1991).

Conclusion

The clinician should be aware of mandibular AVM before performing a biopsy, which may lead to torrential hemorrhage and even death of the patient. A preliminary CT angiography has an 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 gold standard diagnostic aid in management of AVM.

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