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Large Arteriovenous Malformation Of Mandible --- A Case Report And Review Of Literature

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Abstract*: Arteriovenous malformations (AVMs) of the jaws are extremely rare lesions, which are probably hamartomas or developmental malformations. In this article we present an extremely high-flow AVM of the lower jaw (rt side)with spontaneous exsanguinating bleeding from the intraoral lesion site. Digital subtraction angiography revealed a high-flow, high-shunt AVM. Embolization cannot be performed due to high flow, we did bilateral external carotid artery ligation preoperatively, & segmental mandibulectomy with disarticulation of right condyle. Primary closure was done without any immediate reconstruction.*
KeyWords: *arteriovenous malformation(avm), high flow, angiography, embolization, ECA ligation, segmental mandibulectomy*

I. Introduction

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 . 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 involute over 5 or 6 years .[1] Arteriovenous malformations (AVMs) in contrast to the abnormal proliferation of endothelial cells of a haemangioma, display progressive ectasia of abnormal vessels, lined by flat endothelium. AVMs occur as a result of errors in embryogenesis, are always present at birth, may manifest at any time during life and grow proportionately with the child . AVMs can be subdivided based on the rate of blood flow: "slow flow" (capillary, venous, lymphatic or mixed) and "fast flow" (arterial, arteriovenous, fistulae or shunt) subtypes.[1] 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. About 50% of all bone involvement occur in the skull and the maxillo-facial area.[1]

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. However, the clinical course of malformations involving the mandible and the maxilla remains unclear and unpredictable.[1] Mandibular AVMs are usually treated by surgery and/or transarterial embolization. Surgical treatment consists of wide resection of the mandible, which is difficult and potentially hazardous due to significant blood loss during surgery. Selective transarterial embolization is beneficial in reducing shunt flow, although it has a high risk of recurrence. Therefore, some authors advocate to radical resection after selective transarterial embolization for treatment of high-flow mandibular AVM.[2]

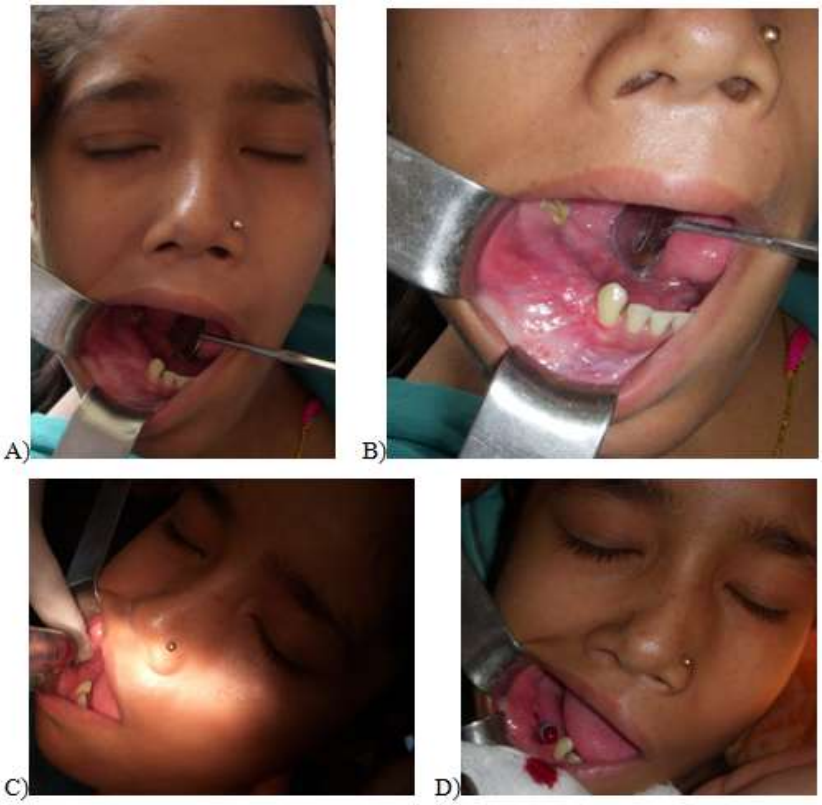
II. Case Report

In September 2014, a 15 yrs old female patient came to the Department of Gurunanak Institute Of Dental Sciences & Research,Kolkata, with a chief complaint of swelling at the right side of mandible for last 4 months. There was occasional pain in that region. On examining the patient, there was a firm swelling involving the right half of mandible from premolar region to right angle of mandible, there was no lip paraesthesia, slightly increased temperature of regional skin, there was expansion of lower border as well as perforation in the lower border. Mouth opening was around 35mm. Intraoral examination revealed there was absence of 44, 45, 46. There was a firm swelling extending from the distal surface of 43 to distal surface of 47. Overlying mucosal surface was lobulated, and there was no surface ulceration, erythema or breach in the continuity. There was marked expansion of both buccal and lingual cortical plate (buccal> lingual) with obliteration of buccal vestibule. On palpation over the expanded area a high flow pulsation was found that was established by the auscultatory bruit heard over the swelling externally. We did aspiration from the swelling from the alveolar ridge region which showed positive blood aspiration. On taking the OPG it revealed that there was a presence of mixed radiolucent and radiopaque lesion with well defined border, with coarse trabeculae with impacted 44, and

45. The anterior and lower extension of the radiolucent-radiopaque lesion could not be delineated properly. This case was initially diagnosed as dentigerous cyst in another institute, but in our opinion it seemed to be a vascular malformation of jaw. Considering our provisional diagnosis we advised the patient to do a contrast enhancing CT scan and CT angiography. The CT scan features included osteolytic expansion of the jaw (cavity like change), expanded space of marrow, and obliteration of trabeculae. These signs were shown well in the bone window setting. Expanded mandibular canal was also found. Numerous cortical perforation found. The lesion crossed the midline and extended upto 32 region.The CT angiography shows extensive vascular lesion of right side of mandible, with nidus of lesion in the right body of the mandible, as well as feeding vessels like engorged right facial artery and lingual artery and also got feeder from left lingual and facial artery. Bilateral ECA angiography was done via right CFA approach. It was reported that there was nidus of serpinginous, tortuous vessels (3.5 x 3 cm) noted in the right submandibular region, with following arterial feeders—lingual, facial, internal maxillary artery, predominately from right lingual and facial artery. Right superficial temporal artery showed partial filling. On the left side it was predominantly from lingual, facial and internal maxillary artery. Draining vein was right internal jugular vein. The impression was AVM (arterio venous malformation) in right submandibular region with arterial feeder from above mentioned branches of bilateral ECA and draining vein being rt internal jugular vein.

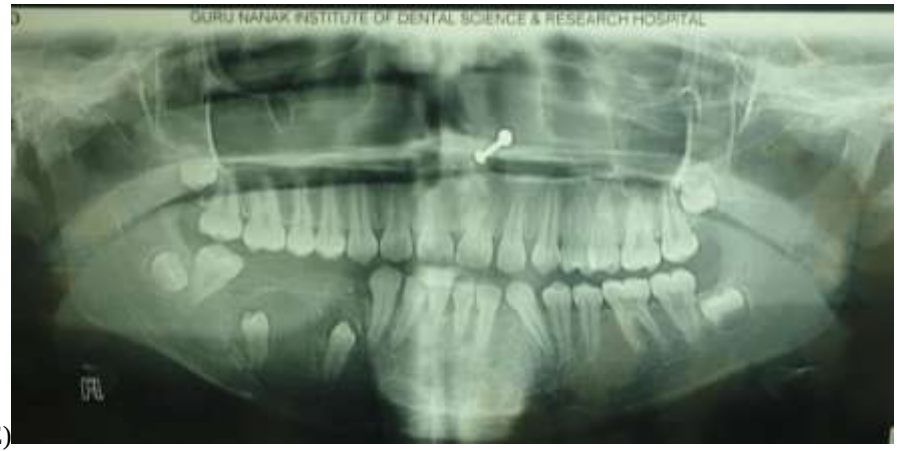
Interventional radiologist interpreted that endovascular embolization was not possible due to high venous flow. It was also interpreted that excision and bilateral arterial ligation can be tried to save the life of the patient. But after all this investigation the patient did not report further. After 6 months the same patient reported to our department with life threatening profuse bleeding from the distal aspect of 47 and the bleeding was continuous for at least 12 hrs. Patient was admitted urgently with a impression compound pack at the bleeding point. Her Hb% and PCV had dropped down to 7.2gm% and 22 respectively. All the other vital signs were stable,2 units of packed cell were transfused preoperatively. Within these 6 months pt visited multiple institute but for unknown reasons, surgical management was not done. We came across with another AP view of skull & mandible which showed total involvement of right half of mandible with condyle.

During peroperative procedure bilateral low level external carotid artery ligation was done, followed by segmental mandibulectomy from 33 with disarticulation of condyle. Peroperatively profuse bleeding was encountered and the patient went into a state of hypovolaemic shock, which was corrected by transfusion of 2 unit whole blood, 500 ml of Haemaccel, bicarbonate was administered along with that ringer lactate and 0.9% NS was given in jet. After the resection primary closure was done. Drain fixed. Post operatively ABG was performed which showed metabolic acidosis, that was corrected further.



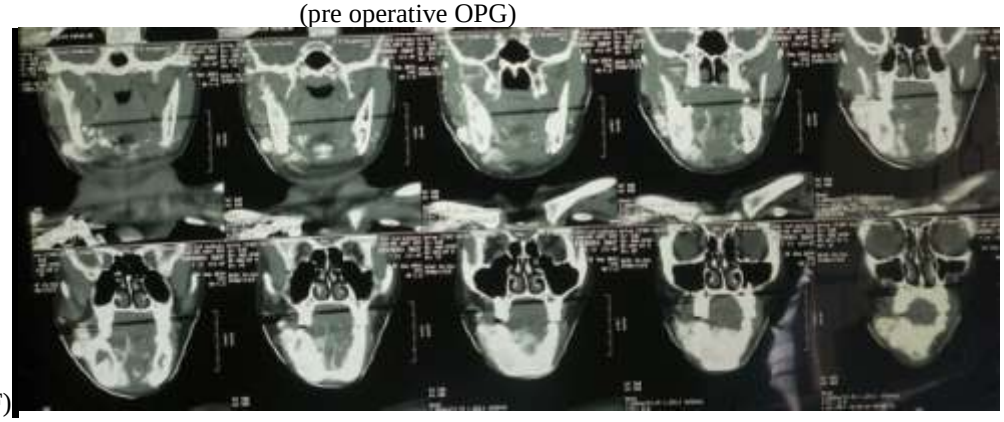
(A+B= pre operative photograph of lesion,C+D= positive aspiration)

E)



(pre operative OPG)

F)



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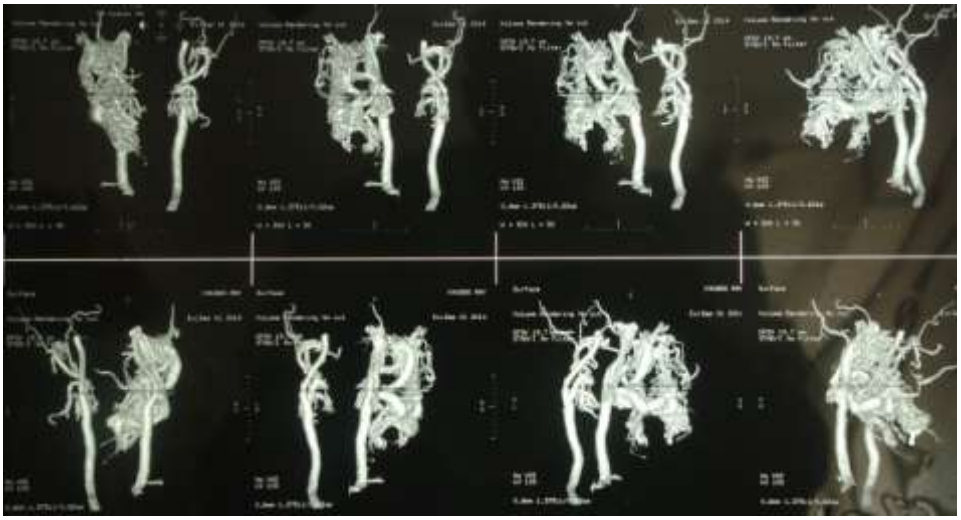
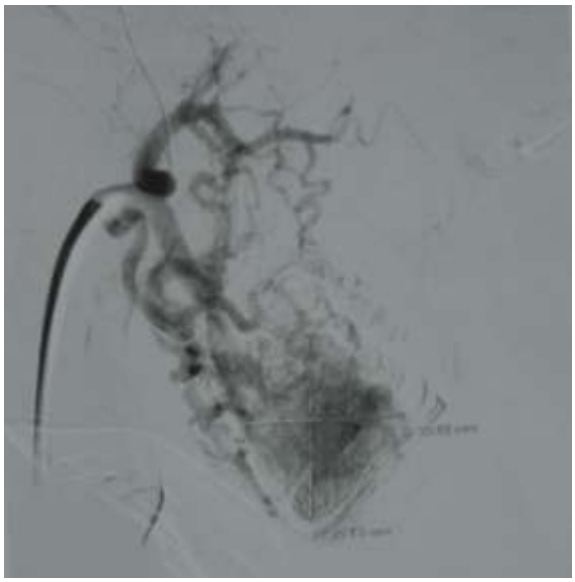
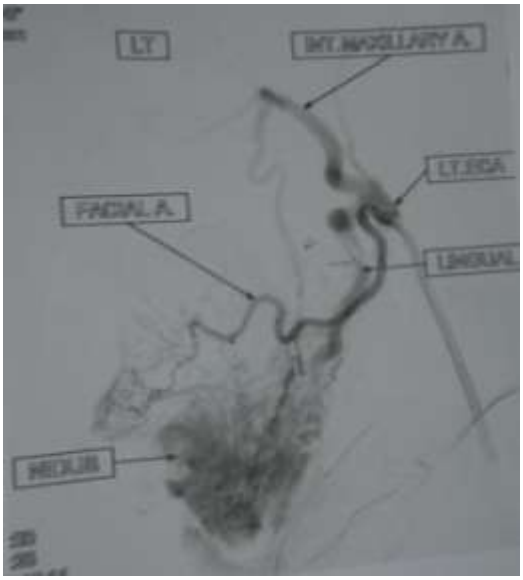


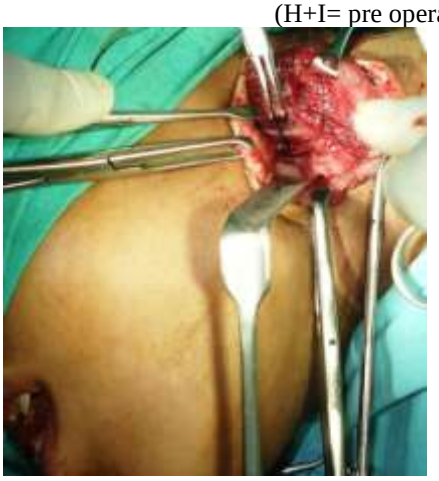
(F+G= pre operative CT scan)

H) I)

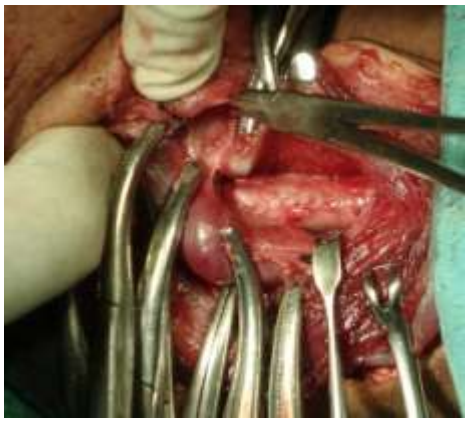




(H+I= pre operative angiography)

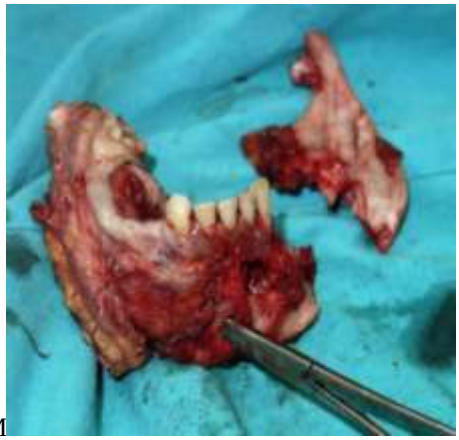
J) 

Left ECA ligation

K) 

right ECA ligation

L) 

M 

(L+M= post operative specimen with perforations)

III. Discussion

AVMs are extremely rare entities that can be life threatening if left untreated due to massive blood loss during tooth extraction or biopsy. Although rare, 50% of all intraosseous AVMs occur in the maxillo-facial region and are extremely infrequent in the mandible.[1]Intraoseous vascular tumors are rare. They are found most often in the vertebra and less frequently in the calvarium but seldom seen in the jaw bones.[3]Very few cases of extraosseous submandibular AVMs have been reported, with 6 cases cited in the literature (5 females and 1

male).[1]High-flow vascular malformations of the jaws are uncommon, and children are mostly commonly involved. The most common presenting complaint is chronic mild intermittent bleeding, which often occurs with minimal oral trauma such as eruption of new teeth or toothbrushing. Facial color and/or temperature asymmetry, audible bruit, palpable thrill, discoloured mucosa, and persistent or recurrent oral infection may also be present. Other complaints include mobile tooth and massive arterial bleeding (especially after dental extraction and eruption of teeth).[4] There are many reports of deaths following dental extraction in undiagnosed cases of AVM of the jaws due to exsanguinating bleeding (Svane et al., 1989).[5] Most are present at birth and enlarge with the growth of the child with no subsequent regression. A few cases may be acquired as a result of trauma.[6]

The panoramic radiograph reveals no pathognomonic features of these lesions . AVMs may have honeycomb radiolucencies or soap bubble appearance and can be interpreted as ameloblastomas, ameloblastic fibromas, odontogenic keratocysts, giant cell granulomas or malignant primary or metastatic tumors.[7]
A CT scan can show the shape, extent and boundaries of lytic expansion of intraosseous AVM, important before a direct intraosseous application of embolisation material.Naturally, a superselective angiogram can provide crucial information on the feeder arteries, draining vein, flow rate and collateral flow of the AVM. These two examinations can be of great help for the definite diagnosis of an AVM.[7]

Management of AVMs in the maxillofacial area is usually complex and requires multidisciplinary team approach. Regarding the treatment of the AVM, many options have been proposed.[7] Observation may be used as a temporary measure in special situations, such as extreme age, pregnancy or refusal of therapy. No spontaneous regressions have been documented. On the other hand, it has been reported that the volume of the lesion may gradually increase.[1]

Arterial ligature was used in the past as a purely symptomatic treatment or before surgery. At present, it is well known that ligation of external carotid artery should not be performed, firstly as many anastomoses promote the rapid appearance of a collateral circulation, and secondly because future embolization would be impossible.[1]

In addition, in our experience, emergency ligation of the external carotid artery is insufficient to stop the haemorrhage because of a large and already developed network of anastomotic blood vessels.[1]
Curettage of the lesion can be curative, but may also lead to excessive bleeding.[7]

Transarterial proximal embolization must be avoided because multiple collateral circulation may develop from the internal carotid artery or vertebral artery and become more difficult to treat. Selective transarterial embolization using several embolic materials has been done. Transarterial embolization with polyvinyl alcohol polymer particles is useful for devascularisation of the AVM, but it has a high risk of recurrence due to recanalization and/or development of collateral arteries to the AVM. In a small number of patients with AVM, including that of the mandibular region, superselective transarterial injection of cyanoaccrylate have obliterated the AVM without recurrence. However, this produces an increased risk for inflammation and necrosis of normal tissue. Some authors have suggested the need for complete or partial resection of the mandible within 24 to 48 hours after diminished flow by transarterial embolization in the treatment of AVM but this may give raise to functional damage as well as cosmetic problems, especially in children. Recently, complete obliteration of the mandibular AVM by direct puncture embolization has been reported. Liquid embolic materials and/or Gianturco coils were introduced into the venous pouch of the AVM through a catheter inserted by transosseous puncture or dental socket.[8]

Permanent embolic obliteration of the malformation requires placement of occlusive material directly into the nidus (core) of the lesion. Even optimal placement of arterial embolic material may fail to fully obliterate the nidus, allowing eventual restoration of flow to the lesion due to arterial recanalization. Under such circumstances it may be possible to obliterate the malformation and control lesional hemorrhage by occlusion of the malformation by direct percutaneous mandibular puncture.[10]

Other concepts include interventional radiology techniques, such as arterial embolisation combined with a surgical approach. Embolisation of feeding vessels may transiently reduce the size of the lesion as collateral vessels may develop within several days. Therefore, embolisation should be used in combination with intraosseous injection of embolisation agents to obliterate permanently the AVM.[7]

Use of a combination of treatment modalities probably is the best way to approach these persistent lesions. By ligation or embolization of the afferent vessels of the lesion prior to surgical curettage or resection, one may reduce the intraoperative hemorrhage. Lowering the blood pressure by positional changes (reverse Trendelenburg) and pharmacologic agents (sodium nitroprusside or deep halothane anesthesia) reduces the intraoperative blood loss and also provides better visualization for the surgical procedure.[9]

However, when resection is required to treat benign lesions in the head and neck (which typically present in children or adolescents) maintenance of facial symmetry, aesthetics, and occlusion are of particular concern. Oka et al. recently described a case where an attempt was made to treat an AVM definitively by resecting the involved part of bone and immediately reconstructing the mandible by using the resected bone as a

crib to carry cancellous bone from the iliac crest and replanting it using a reconstruction plate. Behnia et al. compared transmandibular curettage by proximal osteotomy, and resection and immediate replantation, in the management of AV Mandible found the former to be better for smaller AVM that have not invaded the soft tissues.[11]

Mandibular AVM has traditionally been treated by surgery. Surgical resection generally requires extensive bony resection and complicated reconstruction to try to maintain function of the mandible and prevent disfigurement. Substantial intraoperative hemorrhage may occur, which can be reduced somewhat with preoperative intraarterial embolization.[12]

Surgery is the treatment of choice for AVMs of the mandible. Block resection of the affected area has been suggested, and temporary reconstruction with alloplastic bone plate or with the patient's own free, previously curetted mandibular segment has been reported. More definite bony reconstruction is recommended as soon as possible with a free fibular graft or iliac crest to avoid facial deformities and allow dental implantation. In the past years, some authors have commented that radical surgical resection as in treating cancer. This has been tempered by acknowledgement that radical resection and reconstruction of an extensive benign vascular lesion of the maxillo-facial and mandibular area typically causes severe disfigurement with considerable morbidity. Furthermore, recurrences may occur even after radical resection, and relapses are reported to be even more difficult to treat.[1]

On the other hand, surgical excision of AVMs involving soft tissues such as the submandibular space can be achieved without severe sequelae. No data on relapse have been reported for this very rare condition.[1] Surgery should always be performed after embolization,with a short interval between the procedures, based on the general recommendation that surgery should take place within 48 hours to 2 weeks to avoid revascularization of the lesion.[1]

Resection has been advocated both as primary therapy and as salvage treatment after failure of conservative measures. The indications for resection suggested are obstruction of visual axis; large lesion with thrombocytopenia; obstruction of luminal structures; uncontrollable ulceration, hemorrhage or infection; atypical growth suggesting alternative diagnosis; cardiopulmonary decompensation from arteriovenous shunting and; small lesions that can be excised without cosmetic or functional risk.[13]

Due to the potential danger to the patient in these lesions, surgical intervention has to be the choice of treatment in large lesions refractory to embolisation. Traditional treatment has usually involved the obtaining of proximal and distal vascular control by transfemoral embolisation followed by surgical removal of the lesion, when feasible.[10]

An understanding of the pathophysiology of the lesion has removed ligation of the external carotid and direct surgical approaches from the current armamentarium. The endovascular techniques that replace them are not, however, without risks or complications: many minor problems or major sequelea can occur at the puncture site, along the vascular path and during injection of the occluding material.[15]

But in our case as the endovascular embolization was not possible as depicted by interventional radiologist. So we went for bilateral external carotid artery ligation to reduce the peroperative blood loss and surgical resection was the only hope to save the life of the patient at that condition.

IV. Conclusion

In summary, mandibularAVMshould always be considered in the differential diagnosis of radiolucent lesions in the mandible given the risk of arterial bleeding during dental procedures and biopsy. Currently, these lesions are treated definitively by resection and/or embolization. This example of spontaneous regression raises the possibility that manipulation of angiogenic and trophic factors could be used to help treat high-flowvascular malformations in the future.[14]

Consent: Written informed consent was obtained from the patient for publication of this case report and accompanying images.

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