

Misdiagnosed Arterio-Venous Malformation Causing Massive Bleeding Following Tooth Extraction in A Child: Case Report and Review of The Literature

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ABSTRACT

Arteriovenous malformations (AVMs) are rare, mainly high-flow lesions, comprising a complex network of primitive vessels with interconnecting feeding arteries and draining veins. Their clinical course varies, and while some AVMs are asymptomatic, others cause life-threatening complications. We describe the case of a tooth extraction from the area of misdiagnosed mandibular AVM in a child that caused life-threatening bleeding. After the emergency surgical intervention, the lesion was successfully managed by intravascular embolization

Keywords: vascular malformation, bleeding, embolization, mandible

INTRODUCTION

A comprehensive classification catalog of the International Society for the Study of Vascular Anomalies (ISSVA) divides vascular anomalies into vascular tumors and vascular malformations. Vascular malformations are subdivided into simple and combined capillary, lymphatic, venous, and arteriovenous malformations [1]. The most dangerous type of vascular malformation is an arteriovenous malformation (AVM). They are rare, mainly high-flow lesions, comprising a complex network of primitive vessels with interconnecting feeding arteries and draining veins. The traditional capillary network between the arteries and veins is usually lacking or bypassed. Direct shunts cause a lack of normal down-regulation in pressure and can potentially increase and recruit new vessels. The central section of the abnormal connection and the first section of the adjoining dilated vein is termed the nidus. [2]

AVMs can develop already during fetal life or after birth. They can remain asymptomatic for many years. Initial symptoms may include pain, bleeding, or abnormal growth in the affected area. Objective signs usually include swelling, skin discoloration, warmth, and thrills. Lesions increase in size over time at a variable rate. The clinical course also varies, and while some AVMs are asymptomatic, others cause life-threatening complications. [3]

CASE REPORT

A ten years old girl attended a private dental clinic complaining of repeated episodes of oral bleeding. The dentist performed an orthopantomogram and informed the patient parents that she suffered from an acute dental infection and tooth extraction was necessary. Tooth 47 was extracted under local anesthesia. Immediate massive bleeding followed and was finally controlled by oral gauze packing. The patient was then transferred by ambulance to Farwaniya Hospital where she was admitted to the pediatric ICU. At the time of the admission, the bleeding ceased, but the patient hemoglobin level dropped to 5.8 g/l. A blood transfusion was initiated and consultation by a maxillofacial surgeon on call was requested. At the time of the consultation, there was no active bleeding, and the patient had a gauze pack covering the extraction site.

Based on the history and the photograph of the orthopantomogram (OPG) available on the patient's mother's mobile telephone (Fig. 1), the diagnoses of vascular malformation or highly vascularized malignant tumor were contemplated.

An urgent CT-angiography (CTA) examination was performed and resulted in the diagnosis of arterio-venous malformation of the right mandible (Fig. 2a,b). Another episode of bleeding followed after the return from the CT room, again controlled by an oral pack. Then the patient was transferred to the operating room, where she experienced more severe bleeding before she was orally intubated under dramatic circumstances.

Intraoral examination under GA revealed a gaping extraction wound of tooth 47 filled with pulsating mass profusely bleeding at the slightest touch. The wound was filled with compressed fibrillar oxycellulose secured by a Vicryl 3/0 mattress suture.

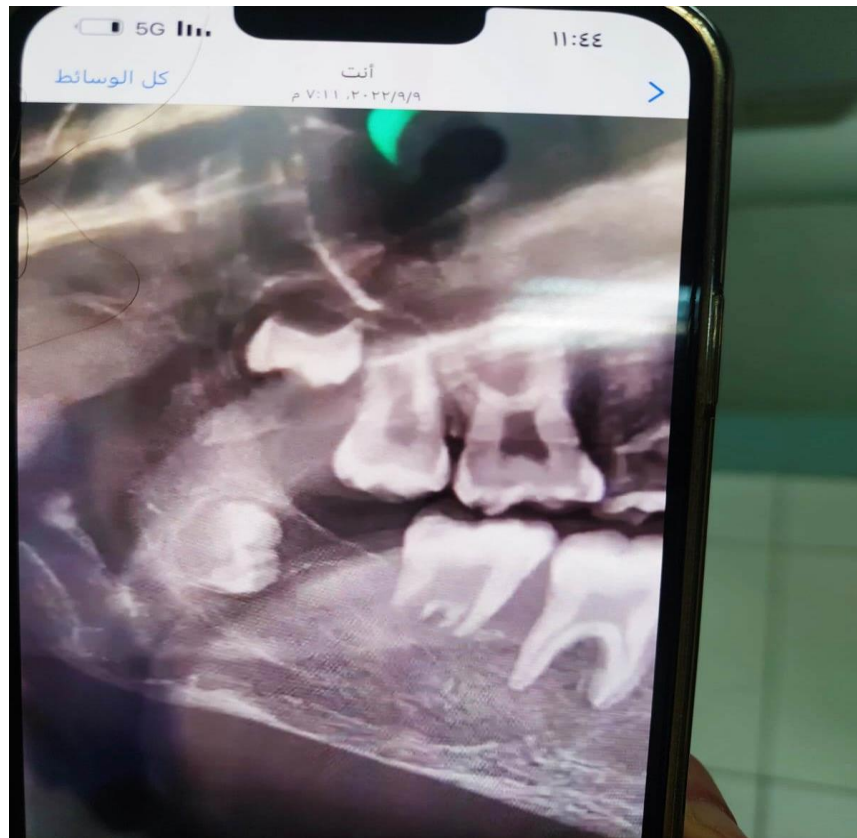


Fig. 1: Part of OPG taken by patient's mother in private clinic before extraction of the tooth 47.



Fig. 2a: 3D CT reconstruction shows non-expanded mandible with discrete cortical resorptions.

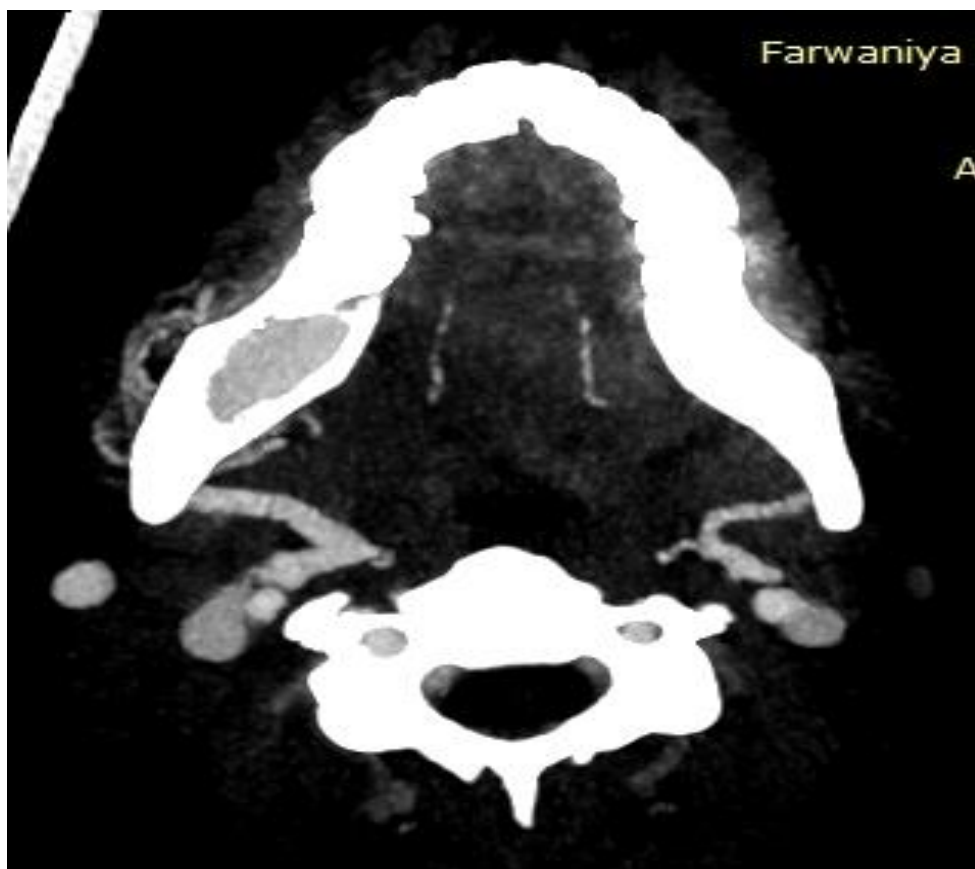


Fig. 2b: CTA (axial MIP reconstruction) – enlarged right ECA. AVM nidus on both aspects of right mandible with dilated venous part inside bone.

The following day the patient was transferred, still intubated to Ibn Sina Hospital for angiography examination and intervention.

Digital subtraction angiography (DSA) study of both internal (ICA) and external (ECA) carotid arteries was done under GA using the right femoral approach and 5F diagnostic and guiding catheter. Right mandibular AVM was detected with arterial supply coming out of right ECA branches: lingual artery, facial artery, and internal maxillary artery. (Fig. 3a). Extensive venous drainage was detected with a venous aneurysm measuring 18 x 8 x 10 mm (Fig. 3b).

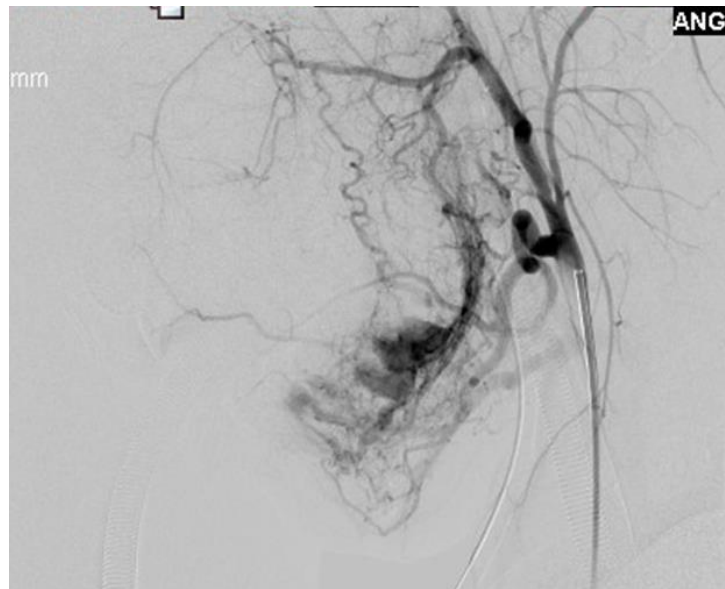


Fig. 3a: Selective right ECA injection. AVM supplied by ECA branches – lingual, facial and internal maxillary arteries.

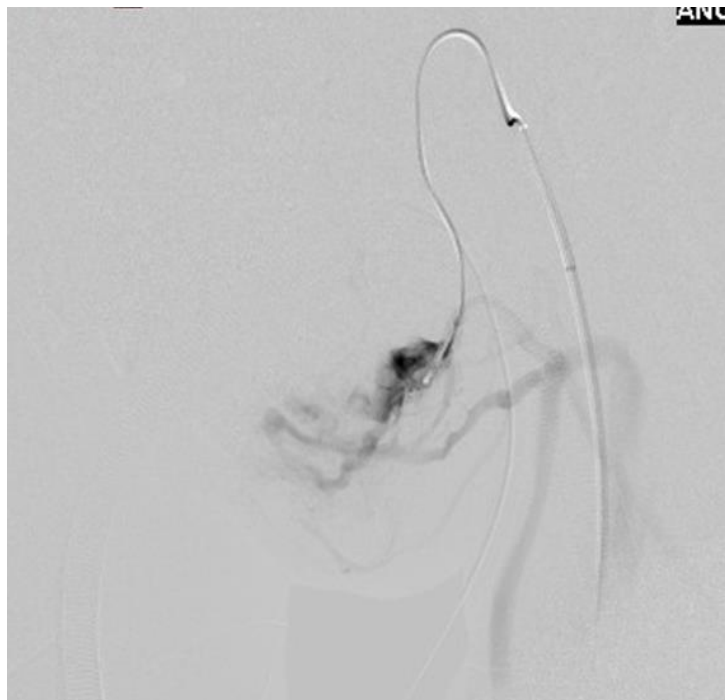


Fig. 3b: Super-selective catheterization of AVM nidus.

Using the coaxial technique, embolization of the main trunks of lingual and facial arteries was done with 4 ml of 500-700 microns spheres (Embosphere, Merit Medical Systems Inc) and single detachable hydro coil (Azur 18, MicroVention Inc, CA). Microcatheter was then introduced into the venous side of AVM through the feeding artery coming from the internal maxillary artery and embolization was done with 3 ml of liquid embolic agent (Onyx 18, eV3 Neurovascular, CA). AVM was completely excluded from circulation (Fig. 4a,b).



Fig. 4a: Right ECA post embolization injection – AVM excluded

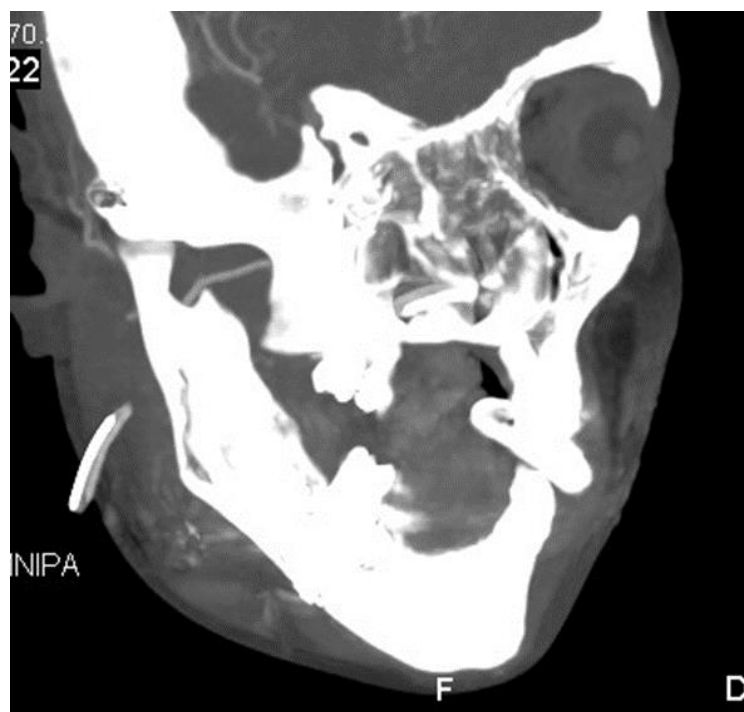


Fig. 4b: F/U CTA – no residual AVM

On return to the Farwaniya Hospital, the patient was observed overnight and no bleeding resumed. She was extubated and transferred to the surgical ward from which she was discharged the following day without any other incident. One week later she attended an outpatient clinic. OPG was done (Fig. 5) and the intraoral suture was removed. She remained under periodic observation. CTA examination was planned after 6 months. However, she visited

the outpatient clinic 6 weeks later with a complaint of sudden swelling in the affected area. Questioning revealed that she was hit to the lower jaw by a sibling in a brawl. Urgently performed CTA yielded a completely normal picture without any signs of the persistent lesion (Fig. 6).



Fig. 5: OPG on follow-up visit one week after the intervention.



Fig. 6: F/U CTA 6 weeks after intervention (VR reconstruction) – no residual AVM

During the following weeks, the patient experienced increasing mobility of tooth 46, growth of granulation tissue, and pus discharge in its vicinity. Tooth 46 was finally extracted under mandibular anesthesia in an outpatient setting. The extraction was followed by abundant bleeding however it was possible to control it by packing the socket with compressed fibrillar oxycellulose. One week later the patient reported no more bleeding.

DISCUSSION

AVMs are congenital lesions attributable either to the persistence of a primitive arteriovenous connection or the development of such a connection after its initial closure during embryonal life. Their prevalence is estimated at 140–500 per 100 000 people (0.14–0.50% of the population). [4]

About 50% of AVMs affect the head and neck with a slight female predominance (1.5:1). During early childhood they usually remain dormant and increase in size during adolescence and adulthood. About 80% are detected during childhood and all will ultimately progress. Hormonal changes during puberty and pregnancy, together with injury have been suggested as potential triggers. Spontaneous regression is almost never seen. [2]

Typical manifestations of AVM include facial swelling and asymmetry, mobility of the associated teeth, pain, skin discoloration, audible or palpable pulsations, and paraesthesia of the lower lip and chin. Hemorrhage can accompany the routine eruption of teeth. [5] Interventions such as dental extraction or biopsy may result in profuse and potentially life-threatening hemorrhage, necessitating emergency management.

AVMs have very few specific features on plain radiographs like OPG. They are usually multilocular lesions with expansion and thinning of the cortex but without perforation. OPG manifestations in the mandible include (1) a clear oval or irregular radiolucency that is mostly centred on the root of the first molar and occasionally with root resorption; (2) an enlarged mandibular canal relative to the contralateral side; (3) a ground-glass appearance with an unclear edge; (4) single or multiple “soap bubble” radio-lucencies; and (5) osteolytic and polycystic changes with signs of crushed and shifted teeth. One or more of these manifestations can be found in all cases. [6,7]

It is considered a wise precaution to obtain a radiograph before the extraction of any tooth [8]. In our case the dentist did it but he completely missed warning features: poorly defined radiolucency with cortical thinning, widening of the mandibular canal, and displacement, respectively premature eruption, of the second permanent molar. These, together with the history of repeated gingival bleeding, should have alarmed him to the imminent danger. However, due to the rarity of AVMs dentists are not familiar with them and life-threatening bleeding following dental extraction or attempted biopsy can be occasionally their first manifestation [5,9-15].

If a tooth is extracted from the AVM region and massive bleeding follows it must be controlled immediately by any available means. Replacement of the tooth into the socket and firmly biting on it is obviously the first choice. Digital pressure over the extraction site [11], ad hoc made thermoplastic plug [5], gauze packing, bone wax, oxycellulose or gel foam can be applied.

Immediate transfer by ambulance to the nearest oral and maxillofacial surgery unit is mandatory.

MRI or CTA are ideal methods to confirm the diagnosis and evaluate the extent of the lesion [5]. The gold standard of diagnosis of AVM is DSA which should be performed by an experienced interventional radiologist and immediately used for endovascular intervention.

Therapeutic approaches for the management of AVMs vary considerably, and no treatment or reporting standards are widely accepted [16]. In past, the accepted management of life-threatening complications of cervicofacial AVMs was ligation of the major feeding vessels. However, rapid enlargement of collateral vessels around the ligature is usually associated with an early return of symptoms. Angiogenesis occurs within weeks to months after ligation. Angiogenetic arteries are predisposed to bleeding and are technically more difficult to penetrate by endovascular methods [17]. Previous ligation of the main feeding vessels prevents catheter access and embolization therapy of the lesion, which is now the preferred treatment for symptomatic AVMs [18, 19].

The surgical management of AVMs is controversial. Partial resection can actually worsen the situation. Any surgery must completely eradicate the lesion while avoiding serious disfigurement [20]. Resection after embolization has become the most accepted surgical treatment. Preoperative embolization can delineate the extent of a lesion and most importantly reduce intraoperative bleeding, [2].

While surgical intervention still has an important role, interventional radiological techniques have offered new methods of control. Monteiro et al. [21] reviewed 50 cases of mandibular AVMs in which embolization was the primary treatment. The materials used separately or in combination were polyvinyl alcohol (PVA), alcoholic solution of Zein, Avitene®, n-butyl cyanoacrylate (NBCA), isobutyl-2-cyanoacrylate, absorbable gelatin sponge (Gelfoam®), ethylene vinyl alcohol (Onyx®), coils, and lyophilized dura mater. Lipiodol® (ethiodized oil) was used as a contrast agent in cases in which the embolization material was not radiopaque. Complete re-ossification was achieved in 12 cases, partial re-ossification in 2 cases, and 25 cases were evaluated as “clinical stability”. Complications we observed in 25 cases, most of them minor and transient.

The use of Onyx® has enabled the partial or complete control of lesions in anatomically challenging areas of the head and neck [3]. Onyx® is a non-adhesive liquid embolic agent composed of ethylene vinyl-alcohol (EVOH) copolymer dissolved in dimethyl sulfoxide (DMSO) and suspended micronized tantalum powder. Onyx® solidifies by precipitating, rather than polymerizing, when in contact with blood. It has an increased flow rate allowing for longer working time, better primary obliteration rates compared with other embolic agents, along with long-term stability [22]. Onyx® leaves a rigid cast that plugs the nidus permanently and has a characteristic appearance on future imaging. Possible complications of its use are local and perivascular inflammation that can potentially damage surrounding healthy tissue and the black stain that tantalum powder causes in the adjacent skin or mucosa.

In summary, AVMs are potentially life-threatening lesions but due to their rarity, most health providers are not familiar with their clinical and radiologic features. Their management remains controversial and mandates close interdisciplinary cooperation.

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