

Management of Sizeable Arterio-Venous Malformation after Infection Post-embolization: An Uncommon Complication of the Common Problem

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ABSTRACT

Arteriovenous malformations are infrequent anomalies that are most commonly present in the head and neck region. Getting a satisfying functional and aesthetic result in large AVM in the craniofacial region is challenging. Most commonly, these lesions are treated by a combination of embolization followed by surgical resection. Infection after embolization is an infrequent complication. We present a large AVM that involved the upper lip, the floor of the mouth, tongue, and masticator space, which got infected after embolization. Management of this patient is presented and discussed, which led to satisfying functional and aesthetic results.

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INTRODUCTION

Vascular anomalies are lesions originating from vascular and lymphatic systems with different histology and clinical behaviour. Failure of early differentiation of embryogenesis leads to hemangioma formation, while vascular malformations are caused by disturbances in the late stages of angiogenesis.¹

The 50% of Arterio-Venous Malformation (AVM) are located in the head and neck area, mainly inside the central nervous system. Extracranially, the most common site is the head and neck, followed by the limbs and trunk.² Sudden changes in AVM size

occur after some triggering event, such as trauma or hormonal changes like puberty.³ Enlargement of these structures occurs due to increased blood flow secondary to dilatation of vascular channels, shunting, and collateral proliferation. Due to direct connection, veins are dilated and tortuous with palpable thrill. These lesions in the face may cause disfigurement and destruction of tissues and compression of vital structures, and life-threatening risks of bleeding occasionally. The risk of bleeding occurs at mucosal surfaces from erosion into malformation due to ulcerations or infections.⁴ The infection has been described as a rare complication of embolization.

Here, we present a case of a large AVM of the lower lip that got infected after embolization. The treatment and surgical intervention are shown, which successfully managed this condition, leading to an acceptable functional and aesthetic result.

CASE REPORT

A 16-year-old patient presented with complaint of rapidly enlarging lower lip with ulcers and diffuse swelling over the left submandibular area and side for the past six months. The patient had a prior history of debulking/excisional surgery at the same site two years back elsewhere. The lower lip started enlarging

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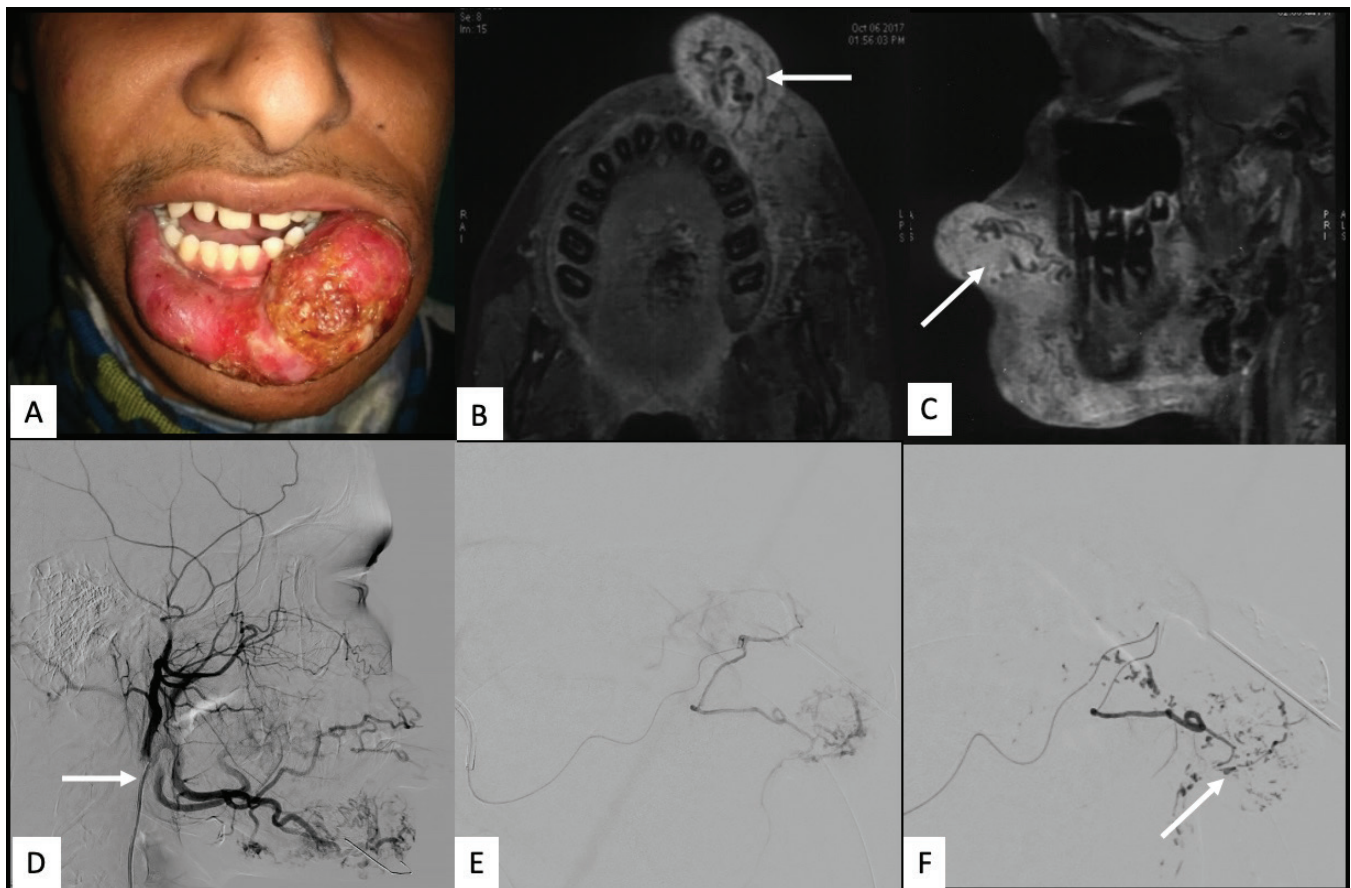


Figure 1(A-E): Clinical image of the patient (A) showing enlargement of lower lip. Magnetic resonance angiography (MRI) T2 fat-suppressed images in axial (B) and sagittal (C) plane showing hyperintense lesion (arrow) with internal flow voids suggestive of arteriovenous malformation (AVM). The digital subtraction angiography (DSA) run taken from the external carotid artery (D) (arrow) showing hypertrophy of facial artery supplying the lesion through an inferior labial branch. The super selective cannulation of the facial artery was done using microcatheter (E). Post embolization presence of glue cast (arrow) within the AVM nidus (F).

after six months of surgery. Later patient developed ulcerations over the lower lip, which were subsequently infected with malodorous discharge. The patient also suffered two episodes of bleeding. The last episode of bleeding occurred 20 days ago. On examination, the lower lip was diffusely enlarged, more on the left half, with infected encrustations with pus discharge. Out-pouting of the lower lip occurred, which caused continuous drooling of saliva (Figure 1A, and 4A). The patient faced severe disability in food intake and maintenance of oral hygiene.

INVESTIGATIONS

Routine investigations, including leucocyte count and pus culture was sent. Culture came positive for *Staphylococcus aureus*, which was sensitive to amoxiclav. Magnetic resonance angiography (MRI) T2 fat-suppressed images in axial (Figure 1B) and sagittal

(Figure 1C) planes showed a hyperintense lesion (arrow) with internal flow voids suggestive of AVM.

The digital subtraction angiography (DSA) run taken from the external carotid artery (Figure 1D) (arrow) showed hypertrophy of the facial artery supplying the lesion through an inferior labial branch.

The post-embolization CT angiography (Figure 2) revealed that the diffuse AVM nidus was located at the tongue, the floor of the mouth, the left facio-maxillary region and extending into the left masticator space.

MANAGEMENT

The patient underwent super-selective embolization of the right facial artery using a microcatheter by n-Butyl-2-cyanoacrylate (0.5 mL) (ENDOCRYL®) (Figure 1E). Post-embolization

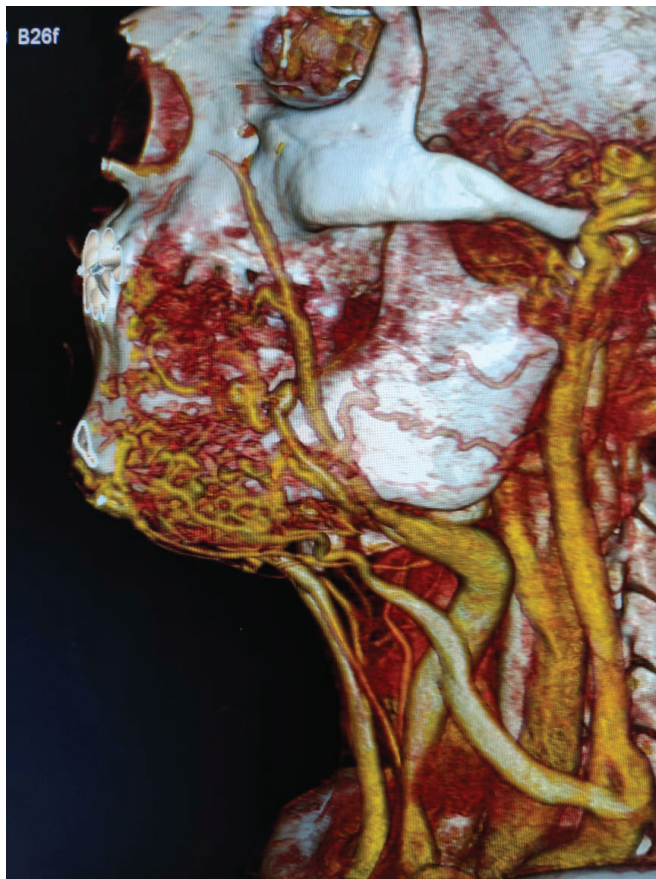


Figure 2: Preoperative 3D CT reconstruction image with enhancement after embolization

presence of glue cast (Figure 1F) confirmed within the AVM nidus. After a few days, the patient experienced sudden enlargement of lower lip swelling and pain. The swelling increased gradually,

followed by malodorous pus discharge from the lower lip. Intravenous Amoxiclav 1.2 gm eight hourly and oral metronidazole 400 mg TDS were started and continued for 10 days, followed by surgery. During operation, the first incision was made on the left side of the neck to expose the feeding vessels. Feeding vessels (branches of left ECA, EJv, and facial vein) were identified and ligated to temporarily suppress lesion's vascularity (Figure 3A). The lower lip's infective part was excised through wedge excision, and the remaining lip was mobilized and advanced. Lip continuity was restored through layered closure. Histopathological examination of the excised specimen confirmed the diagnosis of arteriovenous malformation (Figure 3B). Postoperatively, the patient was managed with intravenous amoxiclav, metronidazole and analgesics. Regular compression dressings were applied and sutures were removed on day 8. At 18 months follow-up, no recurrence of the lesion was reported. The functional and aesthetic outcome was good (Figure 4B).

DISCUSSION

Arteriovenous malformations are uncommon lesions that are most commonly encountered in the head and neck region. The diagnosis of AVM primarily depends on the history and the clinical examination. Radiographic imaging, like MRI or CT helps determine the exact extent, location, and treatment planning. Angiography has become the

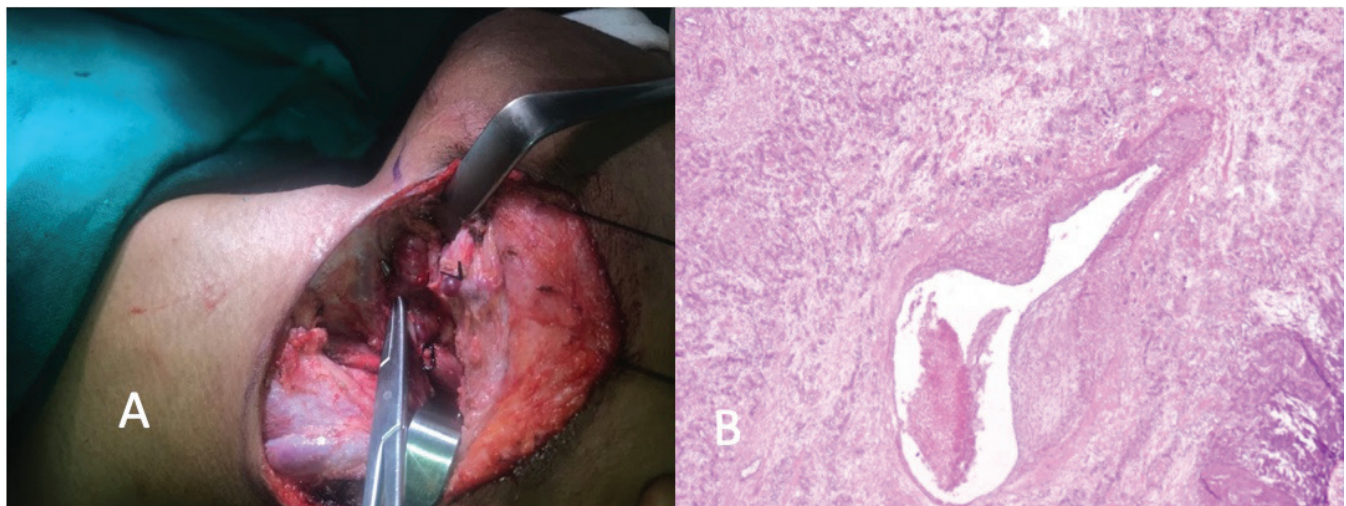


Figure 3: (A) Intra operative image showing ligation of neck feeder vessels. **(B)** Higher magnification H&E (40X) histopathological image showing large feeding vessel with smooth



Figure 4: (A) Pre-operative image showing crustations and slough over huge everted lower lip (B) Post-operative image at 1-year follow up showing satisfactory aesthetic result.

gold standard for AVM diagnosis and management if the embolization is planned. CT angiogram shows tissue enhancement as well as dilated feeding and draining vessels.⁵ MR angiogram is an important diagnostic tool to delineate the nidus and feeders' extent and draining vessels in AVMs. T1 and T2 weighted sequences show typical signal flow voids in high-flow AVMs.⁶

Surgical excision achieves the best result if the lesion is localized. In large lesions, endovascular embolization followed by surgical resection of AVM has been the treatment of choice. Embolization and sclerotherapy have been used to obscure blood flow to AVM, which aids in surgical resection. Embolising agents can be classified into mechanical devices (coils), liquid agents (absolute ethanol, n-butyl

cyanoacrylate), and particulates (polyvinyl alcohol). Complications for glue embolization are localized pain, swelling, erythema, pulmonary embolism, glue mass, muscle dysfunctions, and tissue erosion.⁷

After glue embolization, this patient developed swelling of the lower lip, which later became infected. Infected extracranial AVM and its management have not been reported widely in the literature. Ella *et al.*⁸ reported a case of AVM of the maxilla, complicated by local pathogen infection, which was settled by three weeks of antibiotic treatment. They suggested that infection can be a cause of recurrent AVMs.

In various case series, surgical resection has been performed within 24 to 48 hours of embolization. It has been found that resection during this time period achieves reduced recurrence rates.⁹ With an increased duration between embolization and resection, AVM will expand, and other collaterals may develop. Padreira *et al.*¹⁰ showed no significant difference in the recurrence of AVMs in patients receiving immediate (within 48 hours) or delayed (10.6 days) surgery. They argued that extending the duration is beneficial as it will lead to better delineation of lesion margins with reduced swelling.

In present case, surgical resection was done 14 days after embolization. This delay was required to reduce the infection and associated swelling of the lower lip. There was no difficulty in delineation of margins during the procedure and the post-operative result was satisfactory.

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Consent for publication: Written informed consent for publication of patient's clinical details and/or clinical images was obtained from the patient.

Availability of Data and Material: clinical photographs and case record sheet is available.

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