

A Rare Case: Multiple Mandibular Arteriovenous Malformation with High Output: May a Tooth Extraction Alone Turn to a Catastrophic Condition?

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Abstract

Background: Arteriovenous malformations (AVM) in mandible and neck are potentially life-threatening conditions because of possible massive hemorrhage. They are often first diagnosed by dentists as bleeding. In this case, we report how a tooth extraction turned to a catastrophic condition in a patient with multiple AVMs in the mandibular, submandibular and hyoid region. **Case:** A 19-year-old male patient diagnosed with AVMs in the mandibular and submandibular regions was admitted to our hospital with the complaint of leaky hemorrhage around the lower left 3rd molar tooth. Tooth extraction was planned after embolization procedure. The patient was intubated uneventfully using video laryngoscope. However, an acute bleeding of more than 1600 ml occurred in one minute just after the mouth opener was inserted. The patient was transferred to the Interventional Radiology and other branches of the high-flow AVM were embolized. The patient was taken back to the operating room and the tooth was extracted uneventfully. **Conclusion:** In AVM cases, trauma should be minimized during induction of general anesthesia and intubation should be performed gently with fiberoptic bronchoscopy or video laryngoscopy. We conclude that a multidisciplinary approach as applied to our patient would be valuable in such cases.

Keywords

Arteriovenous Malformations, Tooth Extraction, Anesthetic Management

1. Introduction

Arteriovenous malformations (AVMs) are usually congenital abnormal connec-

tions between arteries and veins. AVMs in the mandible and neck region are rare and potentially life-threatening conditions because of possible massive bleeding [1] [2]. Due to its uncertain clinical and radiographic presentations, AVM is first diagnosed usually by a dentist as bleeding. Endovascular embolization can effectively treat most lesions with limited tissue involvement. However, in selected cases, surgery (mandible resection) can be performed together as part of the treatment [1] [2]. In this presentation, we report how a simple tooth extraction turned into a catastrophic situation in a 19-year-old male patient with multiple AVMs in the mandible, submandible and hyoid regions, in the light of the relevant literature.

2. Case Report

A 19-year-old, 73-kg male patient diagnosed with AVMs in the mandibular and submandibular regions according to the test results performed after a serious hemorrhage following first tooth extraction at age 6, was admitted to our hospital with the complaint of leaky hemorrhage around the lower left 3rd molar tooth. Previously, four sessions of embolizations were performed in an external center. Preoperative blood tests revealed a hemoglobin value of 6.2. Two units of erythrocyte suspension were applied. In CT Angiography, it was determined that the cause of the bleeding was the AVM located just below the tooth. Embolization was performed twice by Interventional Radiology. Tooth extraction was planned after embolization procedure because hemorrhage was triggered due to the movement of the tooth. It was observed that the patient, who was taken to the operating room, continued bleeding slightly from the root of the tooth. Routine ASA monitoring was performed. The patient was intubated with 1 mg/kg lidocaine, 2.5 mg/kg propofol and 1 mg/kg rocuronium using rapid serial intubation method, with a videolaryngoscope, protecting the bleeding area. Before the procedure, two large venous accesses were obtained in case of bleeding and a left radial arterial cannula was inserted. In the beginning of the surgical procedure a projectile hemorrhage was observed while the Brown Davis mouth opener was placed. An acute bleeding of more than 1600 ml occurred in one minute. Right femoral catheter and urinary catheter were inserted immediately. Hemodynamic parameters were tried to be maintained with 1000 ml of colloid and crystalloid fluids until the blood products arrived. Two units of erythrocyte suspension and one unit of FFP were applied. When the bleeding area was under control and stable hemodynamics was achieved, assistance was requested from the Interventional Radiology Unit. The patient was transferred to the Interventional Radiology Unit immediately and it was observed that the patient's AVM was not fed from a single site, but also from the ophthalmic and lingual arteries, therefore the bleeding area could not be fully embolized beforehand. A few more branches were embolized and the flow rate of the high flow AVM was reduced (**Figure 1**). During the interventional procedure, 2 more units of erythrocyte suspension and 1 unit of FFP were applied. The patient was taken back to the operating

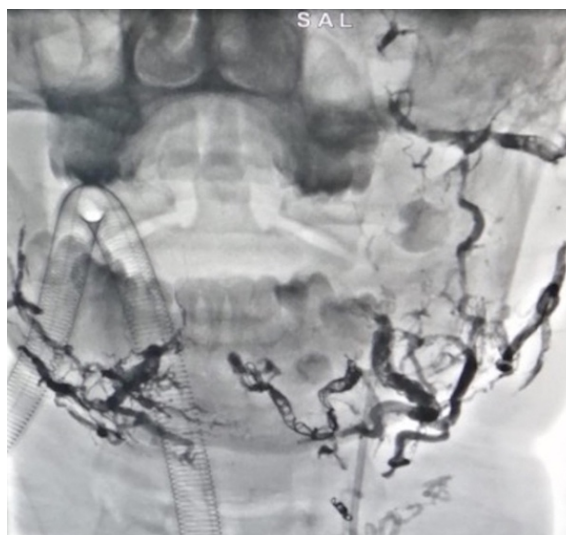


Figure 1. Angiography of embolizations in the submandibular regions.

room and the tooth buried in the mandible was removed with very little bleeding due to a decrease in pressure. Bleeding area was sutured and a liquid embolizing agent was applied. The total amount of bleeding was 2200 ml at the end of the case. The patient with stable hemodynamics and sufficient urine output, was taken to the PACU. After a 12-hour hemodynamic stable period he was extubated and transferred to the service. Informed consent was obtained from the patient for the presentation.

3. Discussion

AVMs are benign lesions with abnormal connections of arteries and veins. They may arise in any part of the body and can rarely be found in bones and jaws [3] [4]. AVMs of the mandible constitute 0.5% - 1% of all lesions [5]. Intraosseous AVMs especially have an increased life-threatening risk of hemorrhage. Tooth extraction or biopsy can cause major blood loss. The complex morphology and the presence of teeth make the AVMs of mandible more difficult to identify than other intraosseous AVMs [6]. Symptoms can range from maxillofacial asymmetry to severe bleeding. CT, MRI and MRA may localize the arterio-venous shunt lesion, but an arteriography is essential for diagnosis and treatment plan [2].

Management of AVMs is usually complex and a multidisciplinary approach is required for a successful outcome. Treatment of AVMs includes removal of the lesion, hemorrhage control during surgery and prevention of recurrence. Radiotherapy may only be a treatment option for inaccessible AVMs. Any deformity of the bone can be corrected surgically [7]. At present, superselective angiographic embolization is considered first line treatment, alone or in combination with surgical approach to reduce intraoperative bleeding. Occlusion of the lesion is accomplished using movable balloons, coils or liquid glue [2]. For lesions that require resection, radiographic embolization is often performed one to two days before surgery to minimize blood loss [8].

Very few cases of submandibular AVMs are reported in the literature [1] [2] [9]. Although preoperative endovascular embolisation reduces intraoperative blood loss during surgery, when it occurs, it can be rapid, massive and difficult to control. Therefore, proper measures should be taken and blood products should be prepared preoperatively. As in our case, preoperative endovascular embolisation was performed before surgery. In spite of preoperative procedure we had a serious bleeding. Clinicians should keep in mind that there is always a possibility for additional arteries feeding the AVMs and keep in touch with the interventional radiology team intraoperatively.

The basic principles of the anesthetic approach include smooth induction and intubation with tight blood pressure control. Compared to embolisation, surgery has an intense stimulation with a possibility of sudden massive blood loss. Anesthetic management should be directed to suppress the noxious stimuli and evaluate the volume status. The hemodynamic response to laryngoscopy, intubation and skin incision must be anticipated and the appropriate anesthetic drugs should be administered at the appropriate time. Monitoring devices are applied before the induction of anesthesia. Arterial catheter should be placed after induction, but before tracheal intubation, for direct measurement of blood pressure. Fiberoptic bronchoscopy or videolaryngoscopy are beneficial since they allow for a gentle intubation [10]. As in our case, anesthetic management was performed successfully and blood products were applied immediately.

4. Conclusion

It is important that blood products are prepared beforehand due to the possibility of sudden and severe bleeding. Trauma should be minimized during induction of general anesthesia and laryngoscopy, and if possible, intubation should be done gently with a fiberoptic bronchoscopy or a video laryngoscopy. Keeping in mind the fatal complications, PACU or intensive care unit can be planned for postoperative follow-up. We conclude that a multidisciplinary approach (anesthesiology, ENT, interventional radiology, maxillofacial surgery) as applied to our patient would be valuable in such cases. The patient gave informed consent for the publication of his case.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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