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A Life Threatening Bleeding Arteriovenous Malformation of the Maxilla Complicated with an Acute Cerebral Ischemic Stroke Recovered: Case Report and Review of Literature

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ABSTRACT

Arteriovenous malformation (AVM) of the head and neck is a rare and potentially life-threatening entity due to massive hemorrhage, which poses a challenge to surgeons treating patients with these lesions.
The head and neck region accounts for approximately 50% of all arteriovenous malformations (AVM), and the same region also accounts for about approximately 50% of intraosseous AVM in the maxillary facial region, which can be very dangerous, as a simple tooth extraction or a biopsy can lead to a major hemorrhage that may eventually cause death. The Treatment may be surgical or non-surgical.
We present the case of a 27-year-old patient with an AVM of the maxilla complicated during his hospitalization for an acute cerebral ischemic stroke, from which he recovered.

Keywords: *AVM; Embolization; Acute cerebral ischemic stroke.*

Introduction

The area of the head and neck is marked by its highly vascular nature, which can be a real challenge for surgeons when they encounter a vascular lesion. AVMs are congenital malformations of the blood vessels, which can be categorized according to the type of the vessel involved or the hemodynamic features.

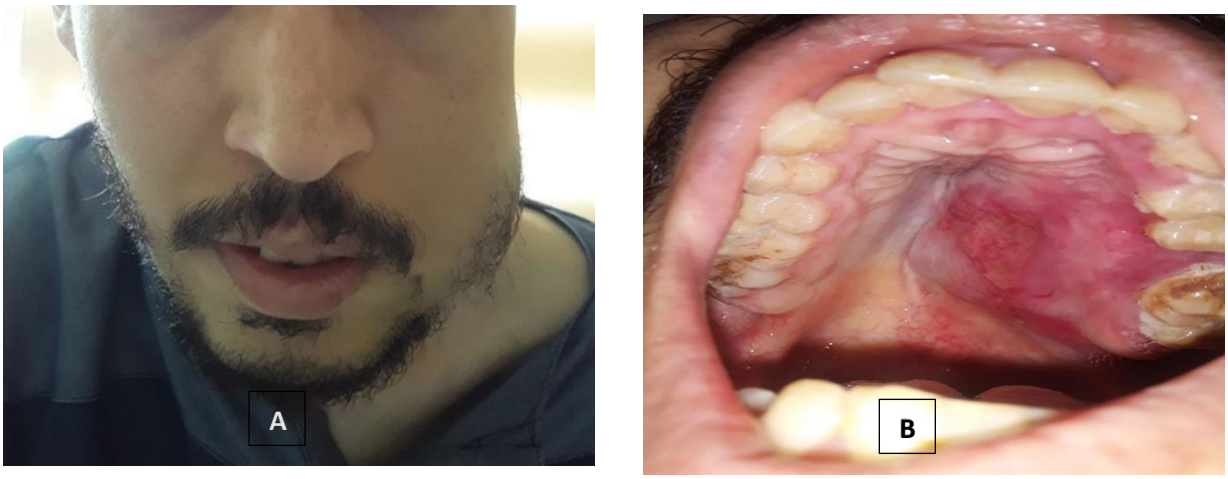
Although they are uncommon in the maxillomandibular region, they can be fatal because of severe tooth bleeding [1]. The age of the patients, as well as the extent, and location of the AVM and many other factors, can all influence the choice of treatment (surgical or non-surgical) that is selected. Surgery can be performed in selected cases in combination with embolization.

In this article, we present a case of maxillary AVM complicated by massive hemorrhage and acute cerebral ischemic stroke.

Case Report

We report the case of a healthy 27-year-old male with a three-year history of a maxillary MAV who had never been treated. The patient was transferred from another hospital to our ENT division because of a massive intraoral hemorrhage. He had been bleeding constantly from his mouth for almost 45 minutes when he was examined in the emergency room, and the flow did not stop despite our efforts to apply direct pressure and topical vasoconstrictors. Physical examination showed a left maxillofacial pulsatile mass (Figure1) associated with a palpable thrill, audible bruit, and massive bleeding from the mouth. His pulse was 125, and blood

pressure was 118/88, and then dropped to 50/34. The hemorrhage was uncontrolled by just the packing, so we took him immediately to the operation room, where we performed a temporary ligation and cauterization of the veins bleeding in the hard palate and filled the mandibular cavity with sheets of oxidized cellulose, which slowed the bleeding (Figure 2).



Figures 1: Shows the left maxillofacial mass (A) that extends in the intra oral (B).

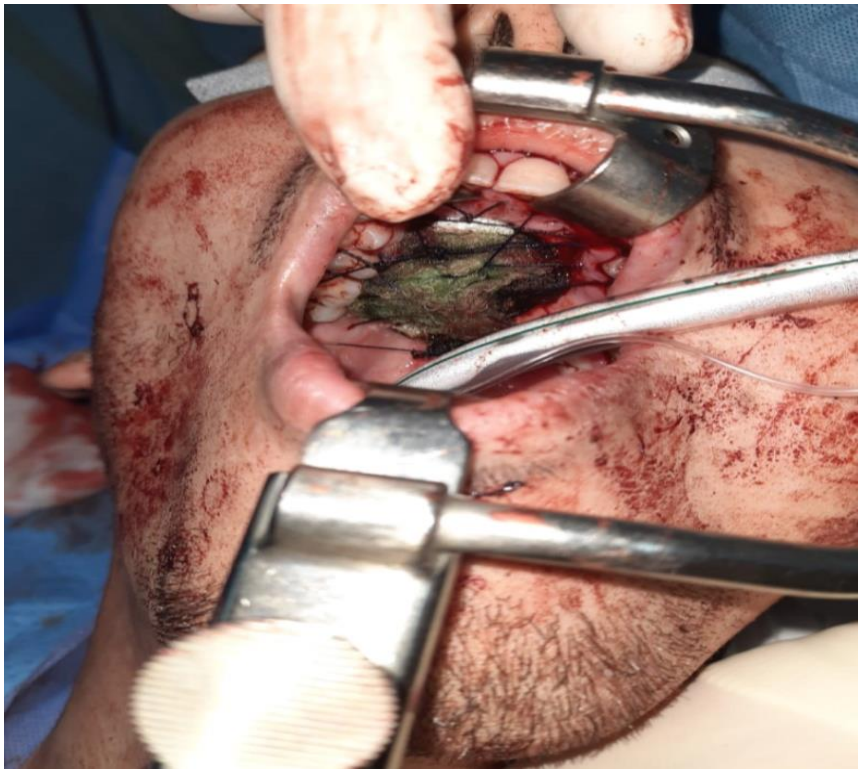
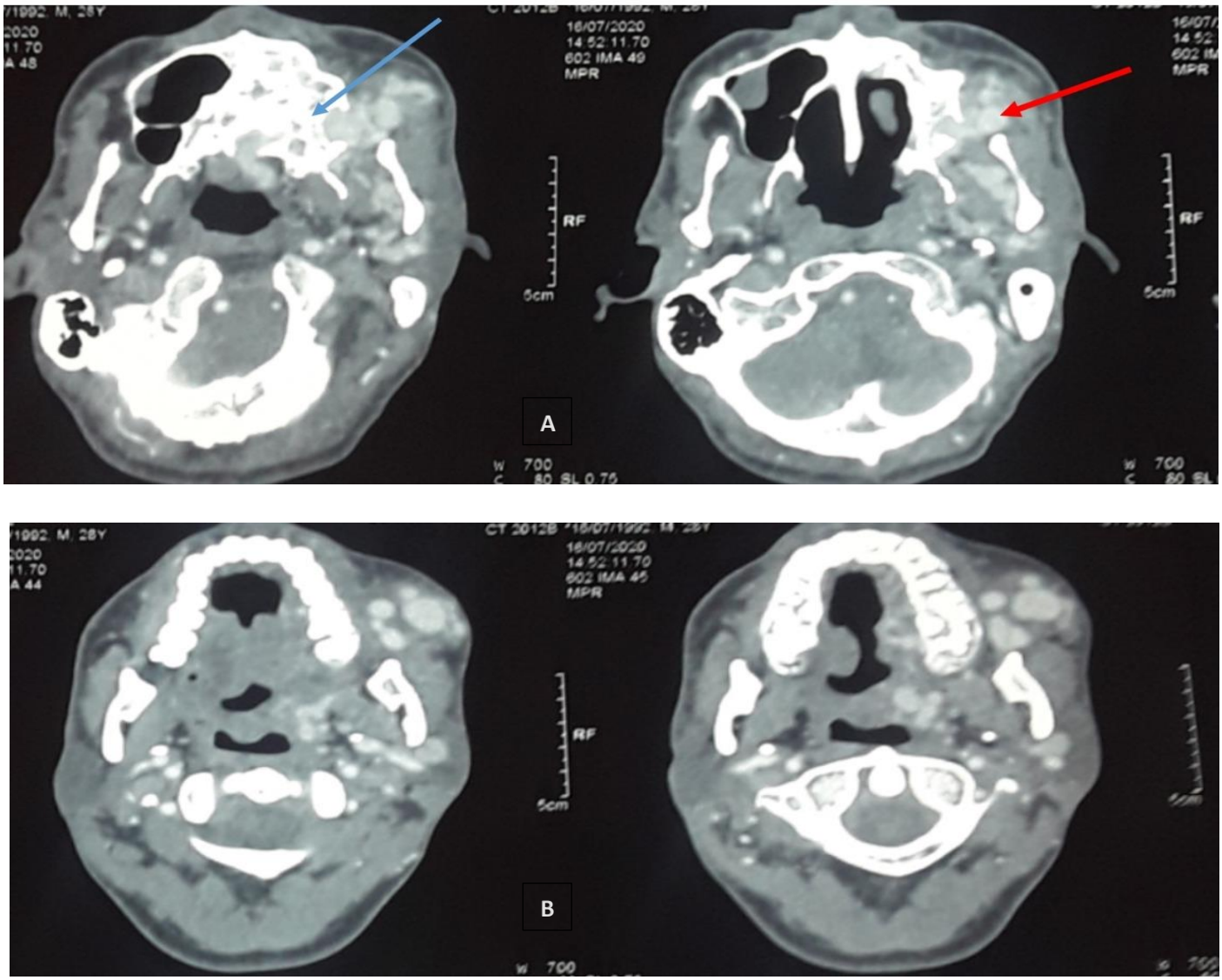


Figure 2: Shows the patient in the operating room after making the packing to stop the major bleeding.

The CT scan showed osteolytic lesions that affected the entire upper jawbone (Figure 3/4). Angiography was performed to confirm the diagnosis of an extensive arteriovenous malformation in the maxilla that was recruited by the internal maxillary artery and the facial artery on both sides and is associated with feeding

vessels from the external carotid and sphenoidal arteries. During angiography, the patient had a severe hemorrhage, and the embolization could not be performed because he was transferred immediately to the ICU. An hour later he developed left hemiplegia, and the magnetic resonance imaging (MRI) revealed an acute cerebral ischemic stroke of the frontal lobe. Two days later, the patient experienced severe bleeding with hypovolemic shock, and severe oxygen desaturation. Immediately thereafter, the patient was transferred to the operating room.



Figures 3: CT Scan showing showed an expansile osteolytic lesion (blue arrow) at the left maxilla bone that is supplied by many collaterals (red arrow) (A, B).

Technique: The patient was placed in the operating room under general hypotensive anesthesia, a tracheostomy was performed to maintain the airway postoperatively, and a large submandibular incision was made. Dissection was done to identify the common carotid artery, and we performed the ligation of the right external carotid. Three other engorged vessels of the left external carotid were attempted because of the extensive anastomotic blood supply, and the hemorrhage was then managed once more. (Figure 5/6)

Subsequently, we discussed the case of this patient in a multidisciplinary meeting, and surgical removal of the

total mass was performed without the preoperative embolization because it was too risky for him.

Technique: Under general hypotensive anesthesia via naso-endotracheal intubation, the patient was prepared, and a Caldwell-Luc incision was performed with dissection, the MAV was identified, and the bleeding was controlled by electrocautery. Osteotomy cuts were performed followed by curettage and hollowing of the maxillary and palatine bones with a rotating handpiece.

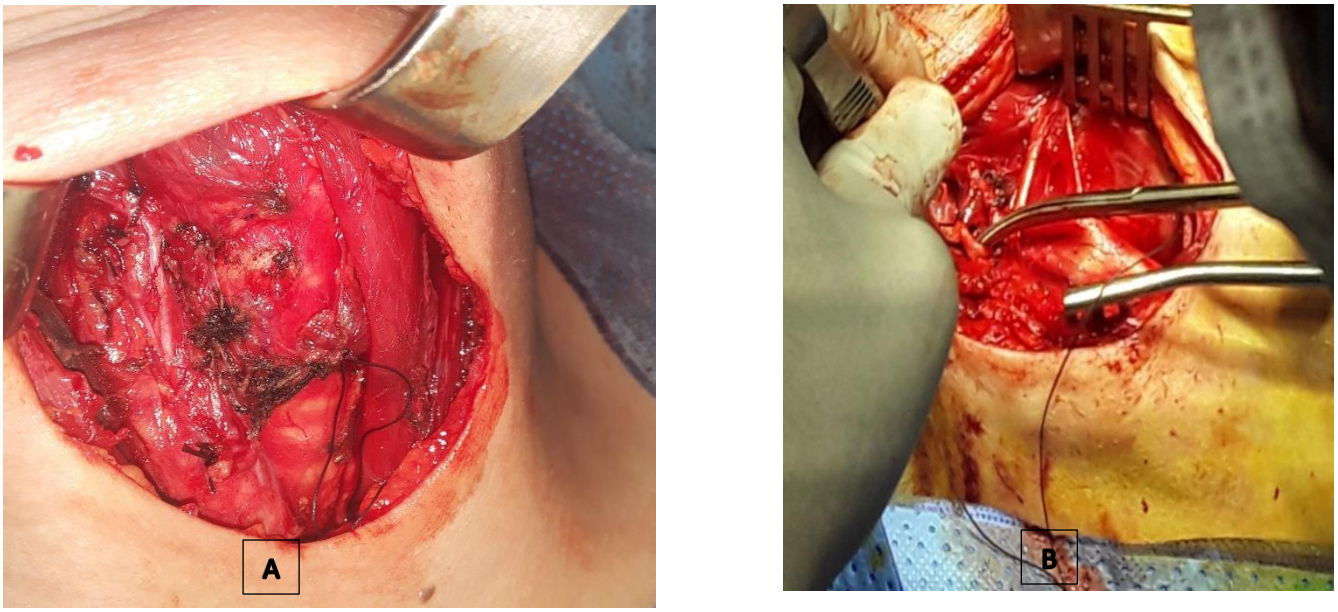


Figure 4: Shows the ligation of the right external carotid (A) and 3 vessels of the left external carotid (B).

The postoperative period was uneventful, the patient recovered fully from the acute cerebral ischemic stroke, and he was discharged on the seventh postoperative day without any signs of bleeding. The final histopathological diagnosis was arteriovenous malformation.

One year later, the patient was presented in the control with no signs of recurrence or bleeding.

Discussion

Arteriovenous malformations (AVMs) are high-flow vascular diseases that frequently involve the mandible and the surrounding soft tissues [2]. They are always present at birth and originate from embryogenesis defects. However, they can also appear at any stage of life (childhood, adolescence, or adulthood) during a tooth extraction with massive bleeding [3].

AVMs may present with non-specific symptoms, including a pulsatile mass, bruits, thrills, and dental loosening, swelling of soft tissues, and changes in the skin and mucosal color. However, hemorrhage is the most common manifestation of these lesions and often occurs around a tooth whose root is buried under a malformation. The resulting bleeding is the most frequent reason for consulting a dentist [4-5].

Radiographically, the MAVs may appear as lytic bone lesions and soft-tissue tumors. They appear as expansile, lytic lesions on computed tomography (« CT scan ») images, and they may also show sclerotic periosteal reactions at the edges of bone involvement. Serpiginous, expansile lesions are observed on magnetic resonance imaging (MRI), and they have signal voids that indicate a high-flow state. Even though a CT scan, or MRI may localize the lesion, super-selective arteriography remains an essential tool for the diagnosis and planning of treatment [6-7]. The management of AVMs usually requires a multidisciplinary team for a good outcome.

The particularity of our case is that the AVM had regressed spontaneously over 3 years, even though no spontaneous regressions have been documented, and then it reappeared spontaneously without any triggering factors. There have been reports of spontaneous regression in both cerebral AVMs and dural AV fistulas, and we found an article describing spontaneous regression of mandibular AVMs for over 12 months and was replaced with trabeculated bone [5]. Another

particularity of our case is that the patient had complications after the hemorrhagic shock with an acute cerebral ischemic stroke, probably because of vessels spasm. Many complications have been recognized in MAVs; however, acute cerebral ischemic stroke is a rare complication that has not been identified in the literature. The type of treatment used, and the outcome were determined based on the location and extent of the vascular malformations. Recently, many articles have suggested various treatments for these AVMs, and the majority of which can be successfully treated using first line super selective arterial embolization, either alone or in combination with surgery, and can effectively cure most of them. According to reports, embolization has a success rate of 70%, which can prevent mutilating surgery. Sclerosing drugs can be injected intra-osseously to lower the number of arterial embolization. Surgery should be saved in cases resistant to endovascular therapy or complications that cannot be treated with alternative treatments (bone fractures or necrosis) [2-7].

However, in our case, embolization could not be performed because of the patient's state (hypovolemic shock, acute cerebral ischemic stroke) and the large diffuse facial AVM with numerous indirect feeding arteries.

Conclusion

In conclusion, AVMs of the maxilla are very aggressive vascular lesions that can cause fatal and life-threatening hemorrhage and require urgent multidisciplinary care for better treatment. Most lesions with minimal tissue involvement can be treated with endovascular therapy. Under certain circumstances, surgery may be combined with embolization.

Additional Information

Consent was obtained or waived by all participants in this study.

Conflicts of Interest

None.

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Financial relationships

All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work.

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