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Case Report

Retrograde transvenous balloon-assisted Onyx embolization of mandibular arteriovenous malformation after hemorrhage

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ABSTRACT

Mandibular arteriovenous malformation (AVM) is a rare lesion, but it often can be presented with life-threatening bleedings. Endovascular treatment of mandibular AVMS has multiple approaches including transarterial embolization, transvenous embolization, direct puncture, and sclerotherapy. In this case study, we present a patient with mandibular AVM complicated by hemorrhage. The patient was treated with transarterial embolization, followed by transvenous sclerotherapy with balloon occlusion of venous outflow. But radical occlusion of AVM was achieved only by transvenous embolization of AVM with Onyx via double lumen balloon, which occluded the venous outflow.

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Introduction

Arteriovenous malformations (AVMs) are pathological vascular anomalies which consist in a direct shunt of blood from the artery to the veins bypassing the normal capillary bed [1]. Approximately 50% of all AVMS are located in the oral and maxillofacial region [2,3]. Mandibular AVMS most often are

an arteriovenous fistulas between the branches of the external carotid artery and the intraosseous mandibular veins [4,5]. There is no exact data on the incidence of mandibular AVMS, but if we extrapolate them on case series, the amount is approximately 1 in 50,000 [6–9]. The clinical manifestations of this disease may include edema of the affected side, small bleeding when brushing teeth, or massive life-threatening bleeding [10–12]. During physical examination, the following

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symptoms can be identified: mobile dentition, pathological deformity of the shape of the mandibular arch, edema of the affected area, gingival injections or bleeding, and bruit during auscultation [4]. Computed tomography angiography and magnetic resonance imaging can be used for diagnosis of mandibular AVM [13]. Selective digital subtraction angiography is necessary to make a detailed analysis of the anatomy and angioarchitecture of the lesion and to choose the most effective treatment strategy. Treatment is aimed to prevent the risks of catastrophic hemorrhages. The goal of treatment is a complete obliteration of AVM. Endovascular treatment may include transarterial or transvenous embolization and sclerotherapy. Embolization can also be performed urgently as a life-saving palliative care before the definitive radical treatment [12,14–16].

Case report

A 36-year-old man was presented with discomfort in the retromolar region and episodes of massive hemorrhage from the oral cavity. Medical history of the patient includes cirrhosis of the liver, the patient got infected in childhood, splenomegaly, and coagulopathy. The patient did not present active complaints of these diseases. The only therapy was performed in 1991, when the splenorenal shunt was inserted; after it the patient no longer received any specific therapy. These diseases could hardly provoke the patient's pathological bleeding. According to the anamnesis morbi, the first bleeding was noted 7 years ago, after 3.8 tooth extraction. But at that time, the patient did not consult a doctor. In 2016, the bleeding became periodic and did not have any obvious cause. Computer tomography showed focus of bone tissue destruction in the region of the branch and the angle of the left mandible. Ultrasound investigation showed vascular malformation in the retromolar region. After another bleeding in May 2017, open surgery was performed in another clinic, but AVM wasn't totally removed.

To stop bleeding, the patient was referred to our clinic for life-saving embolization. Physical examination of left retromolar area demonstrated cyanotic region with visible pulsation and bleeding. Palpation of this area was painful and led to heavy bleeding. Selective carotid angiography was immediately performed and showed high-flow hypervascular malformation of the left-retromolar region with a severe arteriovenous shunt (Fig. 1). Blood supply of malformation was dominantly carried out from the left facial artery and to a lesser degree from the small afferents of internal maxillary artery. Drainage of malformation was carried out through the retromandibular vein.

Vascular access was obtained via 6 Fr introducer placed in right common femoral artery. 4 Fr diagnostic catheter was placed into the left external carotid artery and diagnostic carotid angiography was performed. Then the left facial artery was selectively catheterized. Embolization of AVM was performed via Embosphere Microspheres 500-700 μm (Merit Medical Systems, USA) and particles of polyvinyl alcohol Contour 500-710 μm (Boston Scientific Corporation, USA).



Fig. 1 – Preoperative carotid angiography. (A) External carotid angiogram shows high-flow hypervascular malformation of the left retromolar region. (B) Selective angiography from the left facial artery shows high flow arteriovenous malformation (in the circle), drainage of malformation was carried out through the retromandibular vein (arrowhead).

Remnant afferents from left internal maxillary artery were superselectively catheterized via 2.8 Fr 130-cm Progreat microcatheter (Terumo, Somerset, NJ, USA). Embolization was performed with particles of polyvinyl alcohol Contour 500-710 μm (Boston Scientific Corporation, USA). Follow-up angiography showed dramatic reduction of AVM flow (Fig. 2). After the procedure, bleeding stopped. Five days after the procedure, the patient was discharged from the hospital, without recurrence of hemorrhage.

After 13 days, the patient experienced profuse bleeding which did not stop. The patient was urgently hospitalized

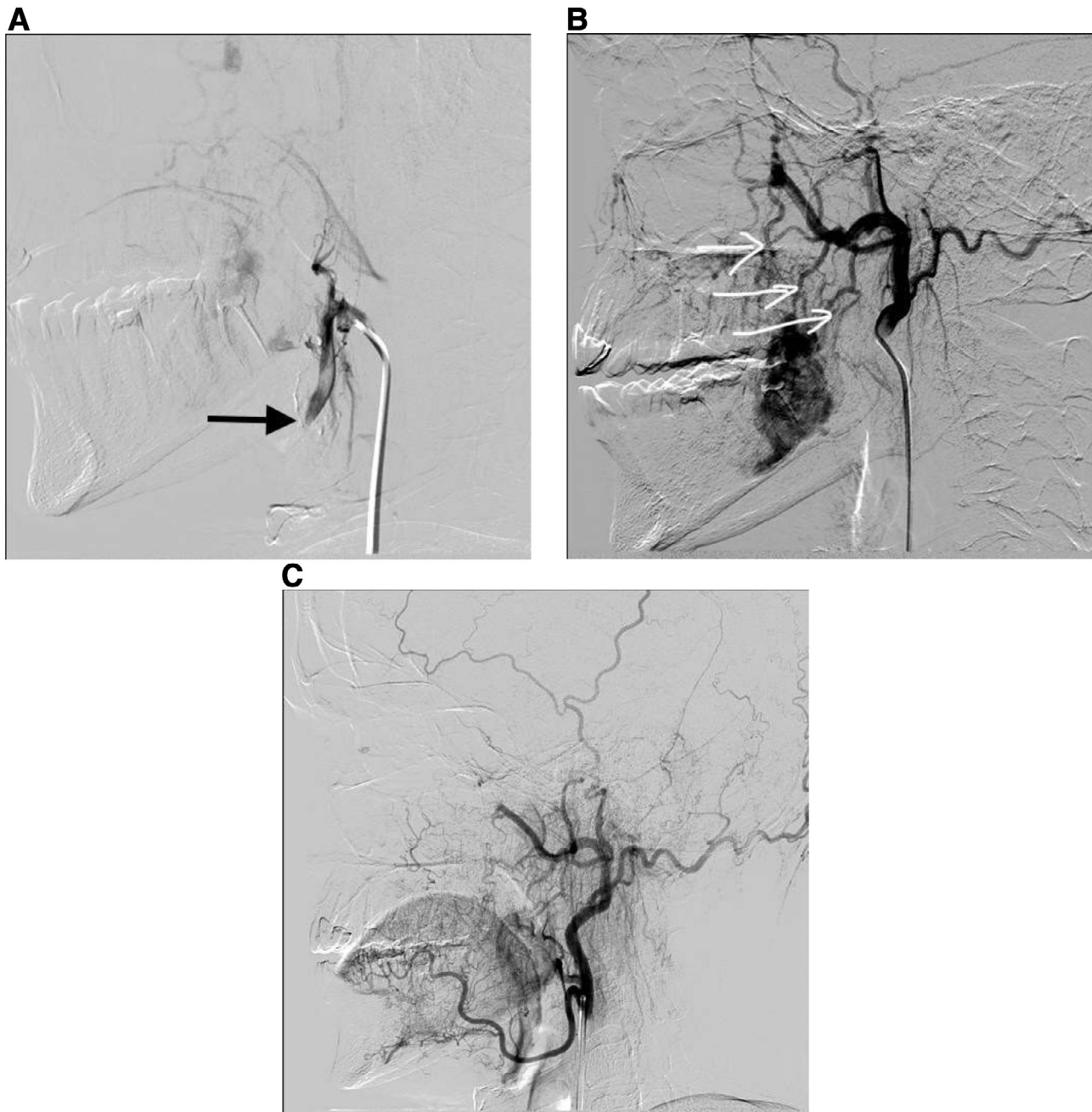


Fig. 2 – Angiographic result after the first embolization. (A) Total occlusion of the facial artery - dominant afferent of AVM (black arrow). (B) Remnant afferents from left internal maxillary artery (white arrows). (C) Subtotal embolization of AVM on the final angiography.

in our clinic for treatment. Selective carotid angiography showed partially embolized AVM of left retromolar region. Arterial blood supply was performed via multiple small vessels from different branches of left external carotid artery and without large afferent, because it was embolized during the last procedure. Venous component of AVM consisted of venous cyst in the retromolar region which was drained via 1 collector-retromandibular vein. All available afferents were selectively catheterized and embolized by particles Contour 300-510 μm (Boston Scientific Corporation, USA). Angiography showed significant reduction in blood supply of malfor-

mation, but pathological arteriovenous shunt was preserved. Since transarterial embolization exhausted itself, it was decided to treat the pathological venous filling with sclerotherapy. The microcatheter headway 21 (MicroVention Inc., USA) was placed in the region of the venous cyst of malformation through the retromandibular vein. Balloon catheter Scepter 4 mm (MicroVention Inc., USA) was placed retrogradely in the area of a single draining venous collector. Balloon catheter was inflated in the draining vein, to achieve a stasis of blood for the performance of adequate sclerotherapy. Sclerotherapy of venous cyst with aethoxysklerol (3.0 mL)

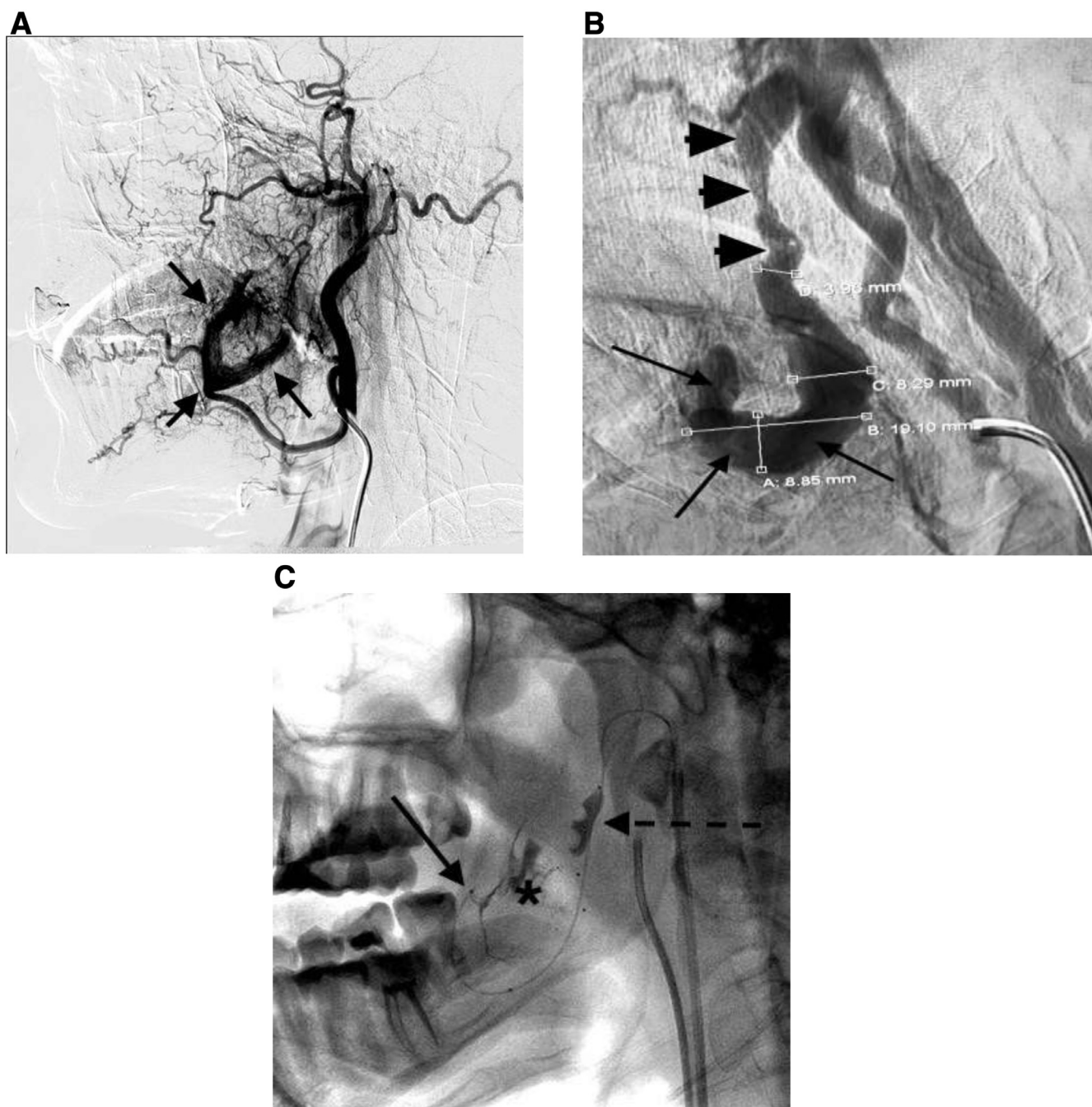


Fig. 3 – Transvenous sclerotherapy of the venous cyst with balloon occlusion of the draining vein (A) Partially embolized AVM of the left retromolar region. Arterial supply of venous component (arrows) was performed via multiple small vessels from different branches of left external carotid artery. (B) Venous component of AVM consisted of venous cyst (arrows) in the retromolar region which was drained via 1 collector-retromandibular vein (arrowheads). (C) The microcatheter headway 21 was placed in the region of the venous cyst of malformation (arrow) through the retromandibular vein. Balloon catheter Scepter 4 mm was inflated in the draining vein (dotted arrow). Injection of the sclerosant in the venous cyst of malformations (asterisk).

was performed with simultaneous finger compression from the oral cavity (Fig 3). Final angiography showed a significant decrease in the rate of arteriovenous blood flow. Simultaneous internal and external compressions were established in order to achieve the best effect of sclerotherapy.

Two days after sclerotherapy, massive bleeding from the oral cavity appeared. Bleeding was stopped only with tight tamponing of the oral cavity. The patient was urgently sent to a catheter laboratory. Carotid angiography showed patent AVM without affordable arterial access for embolization. It

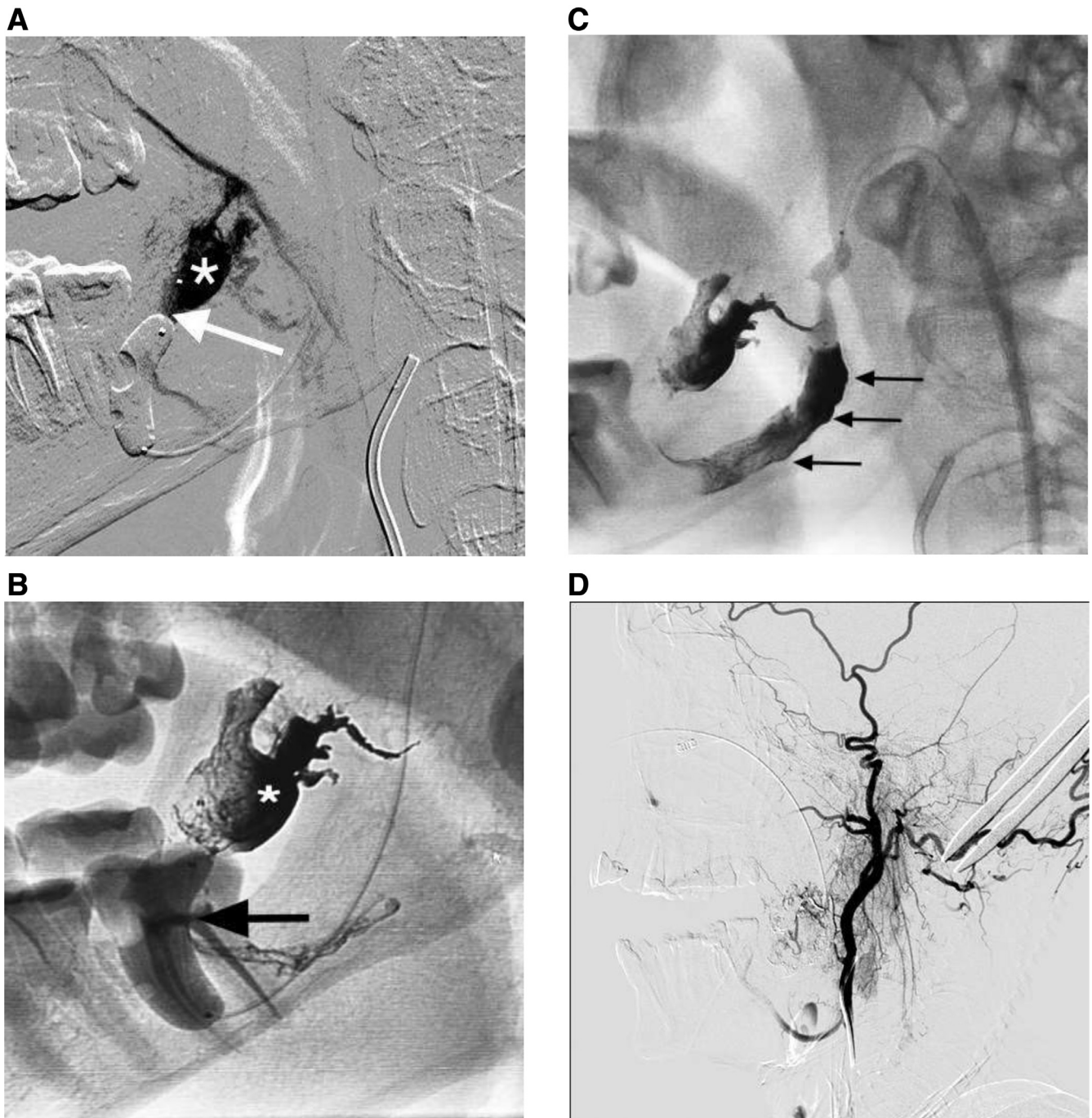


Fig. 4 - Transvenous balloon-assisted onyx embolization of mandibular AVM. (A) Tip of double-lumen balloon (arrow) was placed in the region of the venous cyst of AVM and inflated to perform temporarily occlusion of draining vein. Injection of contrast media through the lumen of the balloon shows stasis of the contrast (asterisk). **(B)** Onyx was injected into the cavity of the AVM cyst (asterisk) through the lumen of the inflated balloon (arrow). **(C)** Draining vein (arrows) was also occluded by onyx. **(D)** Final angiography shows radical occlusion of AVM with complete absence of pathological arteriovenous shunt.

was decided to perform embolization of malformation with Onyx 18 (covidien ev3 medtronic, USA). Due to the lack of adequate arterial access, retrograde transvenous access was selected. A double-lumen balloon Scepter 4 mm was positioned in the region of the venous cyst of the retromolar region through a retromandibular vein. Balloon was inflated

and draining vein was temporarily occluded. In the absence of venous outflow due to the inflated balloon, Onyx was injected (2.5 mL) into the cavity of the AVM cyst through the lumen of the balloon (Fig. 4). Draining vein was also occluded by Onyx. Radical occlusion of AVM with complete absence of pathological arteriovenous shunt was achieved. After remov-

ing the tampons from the oral cavity, the bleeding was completely stopped. Six days after embolization, the patient was discharged in satisfactory condition without recurrence hemorrhage.

The patient came for a repeated examination in 3 months without any episodes of rebleeding. In two years after the operation, telephone monitoring was performed and there was no recurrence of bleeding.

Discussion

AVMs are often localized in the region of a head and a neck. Mandibular AVMs are high-flow malformations. The most dangerous manifestation of the disease is massive bleeding. Conservative pharmacologic therapy is not effective in the treatment of high-flow, intraosseous, mandibular AVMs. Surgical treatment of mandibular AVM (resection with mandibular reconstruction or curettage) is possible, but it often can be complicated by functional or aesthetic deficit [4,13]. For the prevention of massive intraoperative hemorrhage, preoperative embolization is highly recommended [10,11]. The task of endovascular treatment is the radical occlusion of AVM. Embolization can be performed via transarterial or transvenous access and by direct puncture. Radical occlusion of mandibular AVM at most cases can be achieved by transarterial Onyx embolization, and hence, it should be considered as the first choice of treatment. But during urgent embolization or for some other reasons, other embolic agents, such as particles of polyvinyl alcohol or even coils can be used to stop bleeding. It is difficult to achieve a radical occlusion of malformation by transarterial embolization with particles of polyvinyl alcohol and especially with coils. Such partial embolization reduces the degree of arterial supply of malformation, and leads to a hemostasis and to significant decrease of perioperative blood loss during surgery [15,17]. But after incomplete arterial embolization large afferents can be occluded and unavailable for the next stage of embolization that makes later complete occlusion very challenging. During transvenous embolization, incomplete embolization may have more dangerous consequences. In case when the venous outflow is compromised during embolization and the arterial blood supply is preserved, the risk of hemorrhage significantly increases. We consistently experienced both these difficulties in our case. First, after the life-saving transarterial embolization of AVM with polyvinyl alcohol particles, we were able to stop bleeding, but we did not have adequate arterial access to subsequent radical embolization during the next procedure. After incomplete transvenous sclerotherapy of the AVM venous cyst, the venous outflow slowed down, that led to more massive bleeding. And radical occlusion of AVM was achieved only by transvenous Onyx embolization via double lumen balloon, which temporary occludes the venous outflow. For radical treatment of mandibular AVMs, it's usually more effective to use liquid embolic agents such as NBCA and especially Onyx. This case demonstrates the possible technique of mandibular AVMs transvenous embolization, in those cases when, for one reason or another, transarterial access is not available for embolization.

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