

Obliteration of a Large Mandibular Arterio-Venous Malformation with Polymethyl Methacrylate Bone Cement

Case Report

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Abstract

Mandibular arteriovenous malformations (AVMs) are rare and potentially life-threatening vascular lesions. Surgery, embolization, or bone cement implantation is an option for the treatment of mandibular AVMs. We present a case of huge mandibular AVM refractory to multiple embolizations of the supplying arteries, which

was treated with polymethylmethacrylate (PMMA) bone cement implantation after the extraction of a molar tooth.

Keywords: Arteriovenous malformation, mandible, polymethylmethacrylate bone cement

Introduction

Mandibular arteriovenous malformations (AVMs) are rare and potentially life threatening vascular lesions (1). The incidence of vascular anomalies is 1% among the pediatric population, and only 5% is seen in the mandible. Major symptoms are recurrent spontaneous bleedings or serious bleeding after tooth extraction (2). Treatment is unnecessary for small, uncomplicated, and asymptomatic mandibular AVMs. Symptomatic AVMs should be treated with surgery, embolization, or both (3). Preoperative neoadjuvant or curative transarterial, transvenous, or percutaneous embolization and sclerotic material injections may be performed (4-6). We present a case with a huge mandibular AVM refractory to multiple embolizations of the supplying arteries, which was treated with polymethylmethacrylate (PMMA) bone cement implantation after the extraction of a molar tooth.

before bone cement implantation; however, hemostasis could not be achieved without bone cement implantation. Mandibular high-flow AVM nidus was seen on right external carotid artery angiography (Figure 1a). Right internal maxillary artery was preoperatively embolized with 20% N-butyl-2-cyanoacrylate (Glubran; GEM S.r.l., Viareggio, Italy) and lipiodol solution. Residual AVM nidus was seen after the embolization (Figure 1b). The patient did not need blood transfusions throughout the procedure.

The gingival bleeding did not cease after the embolization. Obliteration of the mandibular cavity through the second molar tooth bed with PMMA bone cement implantation was planned. A balloon catheter was placed in the right external carotid artery before tooth extraction for bleeding prophylaxis (Figure 2a). Bone cement was applied to the AVM nidus after tooth extraction. Severe bleeding occurred after tooth extraction and during cement implantation.

It has been seen that the AVM nidus was totally embolized after internal maxillary artery embolization and bone cement implantation on external carotid artery angiography (Figure 2b). Preoperative computed tomography (CT) scans are shown in Figure 3a. The sagittal volume-rendering technique CT image of the patient was shown on Figure 3b, c.

The patient was followed-up for four months, and no bleeding has been occurred so far. The implanted bone cement was trimmed under local anesthesia for a better chewing performance.

Informed consent was taken prior to preparing this manuscript.

Case Presentation

A 12-year-old male patient presented with spontaneous recurrent gingival bleeding due to a huge mandibular AVM. The patient had a history of recurrent gingival bleeding for four years. His complete blood count revealed a slight anemia (hematocrit 33.9%, hemoglobin level 11.6 g/dL) at the time of intervention. Even though the bleeding was excessive during his admission, he did not need prior blood transfusion because of excessive bleeding. The site of bleeding was the right inferior second molar tooth socket. In addition, head and neck examination revealed a 3×3-cm soft swelling in the right ramus of mandible. There was no bruit or thrill. Informed consent was obtained before the initiation of the procedures. Embolization was performed two years ago in our institution, and it was repeated just



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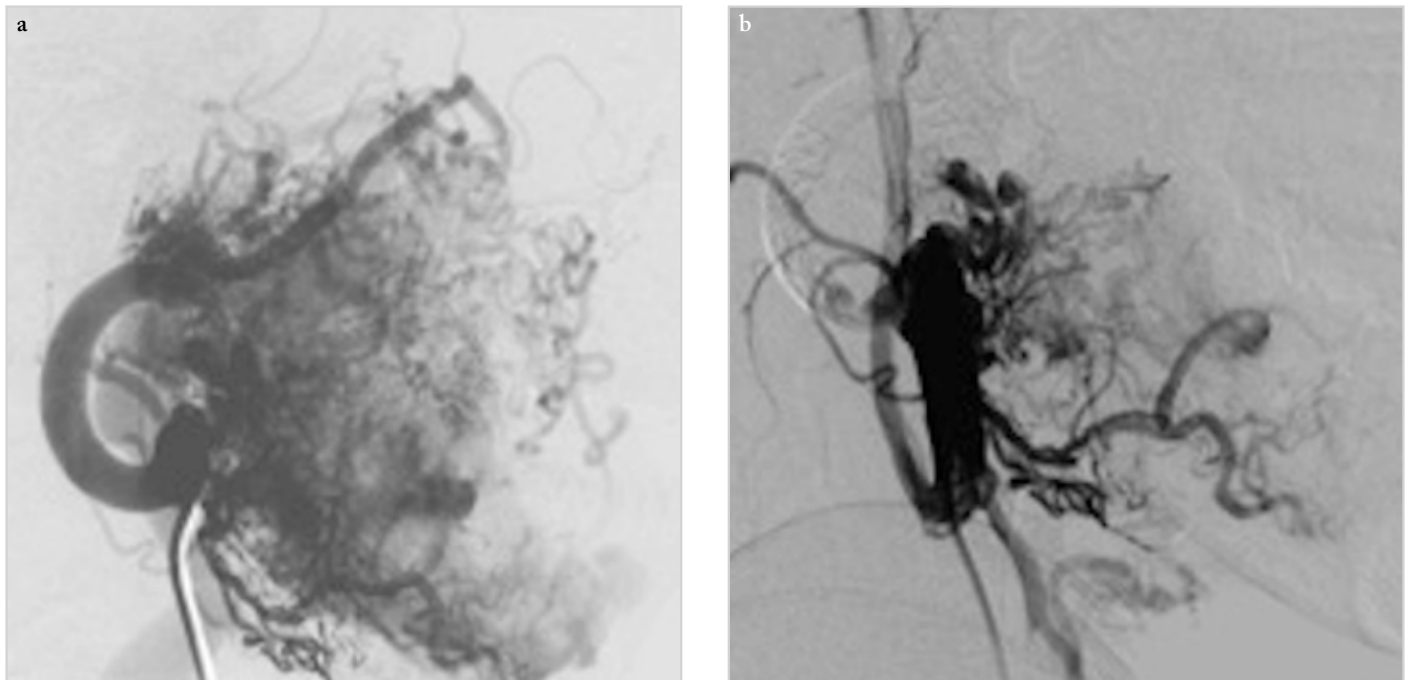


Figure 1. a, b. High-flow mandibular AVM nidus on right external carotid artery angiography (a) Residual AVM nidus after embolization of the right internal maxillary artery with N-butyl-2-cyanoacrylate (b)

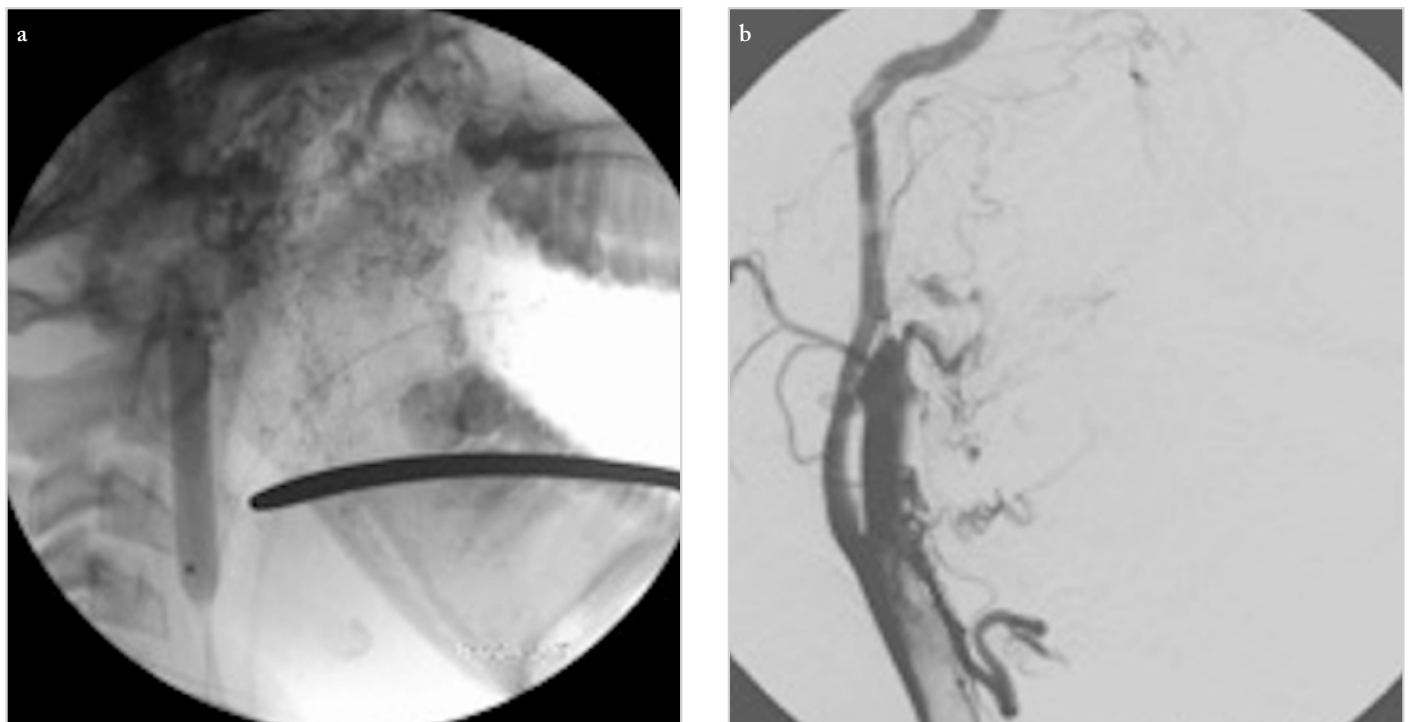


Figure 2. a, b. Placement of a balloon catheter in right external carotid artery before bone cement implantation for bleeding prophylaxis (a) Totally embolized mandibular AVM on right carotid angiography after internal maxillary artery embolization and bone cement implantation to AVM nidus; AVM: arteriovenous malformation (b)

Discussion

Mandibular AVMs are rare and potentially life-threatening vascular lesions (1). These are vascular anomalies characterized by one or more direct or indirect connections without a capillary bed between arteries and veins. They appear because of an anomaly in one or more steps of angiogenesis in the embryo.

AVMs divide into three according to low, high and combined flow (7).

The presenting symptom of our patient was compatible with that of previous literature: spontaneous bleeding and bleeding after tooth extraction (2). A high pressure in the AVM

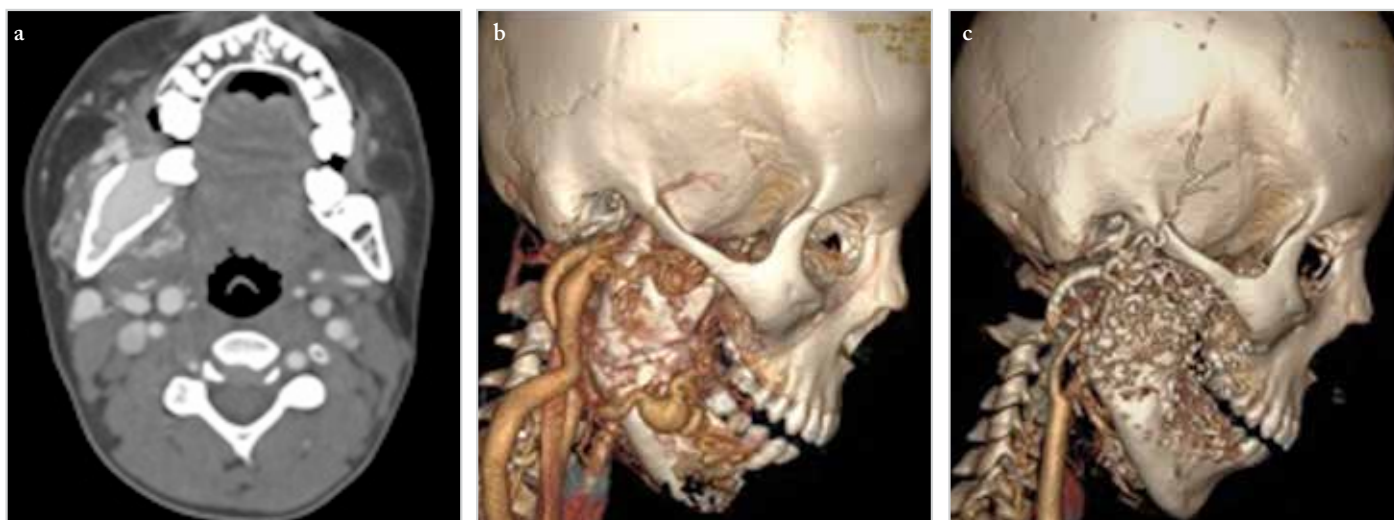


Figure 3. a-c. Preoperative computed tomography showing mandibular AVM (a) Vascular engorgement in AVM nidus and retromandibular and facial veins on preoperative sagittal volume-rendering technique CT image (b) Glue cast because of N-butyl-2-cyanoacrylate (Glubran) is seen on the postoperative sagittal volume-rendering technique CT image; AVM: arteriovenous malformation (c)

nidus causes lytic areas on teeth roots and mobilization of the teeth (5). CT-angiography is very helpful in showing the localization and blood supply of the lesion in the diagnosis of AVMs. CT also shows lytic changes in the mandible and the postoperative opacity of PMMA bone cement as in this case.

Treatment is unnecessary if AVM is small and asymptomatic. Symptomatic cases may be treated with surgical excision and reconstruction; however, the outcome of surgery alone is unsatisfactory and sometimes life-threatening (5). Therefore, preoperative embolization or sclerotic material injections may be used (4-6). Transarterial embolization with polyvinyl alcohol polymers has been found to be effective in the devascularization of AVMs; however, recurrences were relatively frequent because of the growth of terminal arteries (8). In a case with the transarterial superselective application of N-butyl-2-cyanoacrylate was effective and free of recurrences (9). However, there is still concern about recurrence after this treatment. Erroneous mobilization of the injected material to the internal carotid system may cause intracranial complications, that to the ophthalmic artery may cause blindness, and that to the pulmonary circulation may cause even the death of the patient with multiple embolizations after recurrent disease (4-5).

PMMA bone cement is a biocompatible and non-degradable acrylic resin-based implant, which is currently the most extensively used alloplastic material for craniofacial reconstruction (10). PMMA bone cement was successfully used as an occlusive material in an intraosseous arteriovenous lesion (11). Our case is the first in which N-butyl-2-cyanoacrylate embolization and PMMA bone cement implantation was successfully used for a recurrent huge high-flow mandibular AVM in a multidisciplinary manner. Severe bleeding may occur during bone

cement implantation, so blood transfusion should be preoperatively considered. We believe that a multidisciplinary approach with balloon occlusion and combined embolization with cement implantation is curative and the safest method for the treatment of mandibular AVMs.

Informed Consent: Written informed consent was obtained from the patient.

Peer-review: Externally peer-reviewed.

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