Arteriovenous Malformation of the Mandible: Embolization and Direct Injection Therapy

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Arteriovenous malformation (AVM) of the mandible is a rare entity but one that can be potentially fatal as a result of massive hemorrhage. Traditional treatment involved extensive surgical resection of the mandible. With the advent of improved endovascular techniques, interventional radiology is now the best method to control active hemorrhage and ultimately cure these lesions. The authors describe three cases of successfully treated mandibular AVM by percutaneous and/or endovascular techniques.

Index terms: Arteriovenous malformations, mandibular • Embolization • Jaws, abnormalities

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Abbreviation: AVM = arteriovenous malformation

ALTHOUGH arteriovenous malformation (AVM) of the mandible is a rare entity, it must be recognized by dentists, oral surgeons, and radiologists because of its potentially life-threatening complication. A common presentation is a spontaneously loosened tooth that if dislodged or extracted may result in rapid massive hemorrhage and possibly death in an otherwise healthy young patient. The diagnosis of an AVM should be suspected when radiographic imaging demonstrates a lytic lesion in the mandible associated with the loose tooth (1). Traditional treatments included extensive surgical resection and mandibular reconstruction to preserve the obvious functions and esthetics of the mandible. With the advent of modern endovascular and percutaneous techniques, successful treatment of mandibular AVM by interventional radiologic means is possible.

CASE 1

A 15-year-old girl presented with a loose left lower second molar. Investigations performed at another institution included a computed tomographic (CT) scan that showed a lytic lesion of the left mandible and an angiogram that demonstrated a mandibular AVM. Elective preoperative arterial embolization followed by surgical resection were originally planned. While waiting for the procedure, massive hemorrhage began to occur from around the loosened tooth. The bleeding was controlled by extensive packing of the oral cavity. She was stabilized with blood transfusions and transferred to our institution for emergency embolization. The embolization procedure was performed under general anesthesia with transnasal endotracheal intubation because the oral cavity was filled with packing material. Right femoral arterial and venous punctures were performed. An initial left external carotid artery angiogram demonstrated a high-flow mandibular AVM supplied by the internal maxillary (Fig 1a), facial, and lingual arteries. A large venous pouch was present in the left body and ramus of the mandible drained by the retromandibular vein into the left internal jugular vein. The internal maxillary artery was first embolized with large-sized particles (355–500 μm Contour Emboli, Target Therapeutics, Fremont, CA) to decrease the flow rate of the AVM. The facial artery was then catheterized and a venous “road map” was made, allowing transvenous placement of a microcatheter into the venous pouch (Fig 1b). A subsequent synchronous injection of contrast material through the arterial and venous catheters provoked massive rebleeding around the loosened molar (Fig 1c). A 2-mL mixture of 25% glue (Histoacryl; B. Braun, Melsungen, Germany) and 75% lipiodol (Lipiodol Ultra-fluid; Guerbet/Therapex, Montreal, PQ, Canada) was immediately injected through the venous catheter while compression of the internal jugular vein was applied to prevent escape of glue into the systemic venous system. A small residual venous pouch was still present and bleeding persisted, albeit slowed. The oral packing was removed, embolization by direct puncture was performed into the bleeding area around the loose molar with a 18-gauge needle, and a further 4 mL of 50% glue/lipiodol mixture was injected. A postembolization external carotid injection showed complete eradication of the AVM (Figs 1d, e). Repeat angiograms 2 days and 3 months later confirmed continued closure of the AVM. The loosened molar was subsequently extracted along with removal of a small amount of extruded glue material into the oral cavity without any further bleeding complications.
Figure 1. A 15-year-old girl with a left mandibular AVM. (a) Lateral projection digital subtraction angiographic image from an initial left internal maxillary artery injection shows a high-flow AVM with a large intraosseous venous pouch drained by the retromandibular vein. (b) A transfemoral venous microcatheter is positioned with its tip within the venous pouch. (c) Synchronous injection of contrast material through the venous microcatheter and catheter in the facial artery provoked active bleeding into the oral cavity (arrows). (d) Postembolization DSA and (e) unsubtracted images from an external carotid injection show complete obliteration of the AVM. Glue has escaped into the left retromandibular vein (arrow) and a small amount has extruded into the oral cavity at the site of the loose molar (arrowhead).
CASE 2

A 15-year-old boy presented to a dentist with a loose left second lower molar associated with a small amount of bleeding when the tooth was slightly mobilized. A CT study showed a lytic lesion (Fig 2a) and an initial diagnosis of an adamantinoma was made. Biopsy was performed on the lesion in an operating room under general anesthesia, which resulted in massive hemorrhage that could not be controlled with packing. An emergent surgical ligation of the left external carotid artery was then performed, but the bleeding did not stop. Blood transfusions were given and a tracheostomy was made. The bleeding site was packed and the patient underwent angiography, which demonstrated an AVM of the left mandible. Because of ligation of the left external carotid artery, the AVM was now fed by the vertebrobasilar system via collateral with the left occipital artery (Fig 2b). Transarterial embolization was attempted but was unsuccessful because of an inability to navigate a catheter through these newly formed collateral vessels. The patient was then transferred to our institution for treatment. A direct percutaneous puncture into the venous pouch was performed with a bone marrow aspiration/intraosseous infusion 15-gauge needle (Illinois Needle, McGaw Park, IL) under fluoroscopic guidance (Fig 2c). A 2.6-mL mixture of 50% glue/lipiodol (Histoacryl, B. Braun; Lipiodol Ultra-fluid, Guerbet/Therapex) was injected, with near complete filling of the venous pouch, but a small pool of contrast material remained near the second left molar. The packing was removed and the tooth spontaneously dislodged. Active hemorrhage resumed from the alveolar socket. An 18-gauge needle was inserted into the socket and 3.2 mL of 50% glue mixture was injected. The hemorrhage ceased and the AVM became completely obliterated on immediate control angiogram. Follow-up angiograms 1 month and 1 year later showed continued obliteration of the AVM.

CASE 3

A 9-year-old boy reported left lower tooth discomfort. An orthopantogram and subsequent CT demonstrated a well-defined lytic mandibular lesion with erosion of the roots of the first and second molars. A dentist extracted one of the molars in his office and massive hemorrhage occurred. The bleeding was initially controlled with packing so the patient was discharged home from the office. The bleeding resumed later that day. The patient’s father, with use of digital pressure on the bleeding area, was able to gain some control of the hemorrhage and the pair presented to our institution. An angiogram was performed showing a left mandibular AVM. The patient was moved to the operating room and under fluoroscopic control, a direct puncture was made with an 18-gauge needle from the lingual buccal surface. A 3-mL mixture of 33% glue and 66% lipiodol (Histoacryl, B. Braun; Lipiodol Ultra-fluid, Guerbet/Therapex) was injected. Intraoperative angiography initially showed no further filling of the AVM, but when digital pressure was removed, bleeding resumed. The bleeding site was found and a further 2.5 mL of 50% glue mixture was injected and the AVM was obliterated. A control angiogram 3 days later showed continued closure of the AVM and a repeat orthopantogram showed glue material completely filling the venous pouch.

DISCUSSION

Mandibular AVM has traditionally been treated by surgery. Surgical resection generally requires extensive bony resection and complicated reconstruction to try to maintain function of
the mandible and prevent disfigurement. Substantial intraoperative hemorrhage may occur, which can be reduced somewhat with preoperative intraarterial embolization. Radiation has been tried with some success (1,2). Ligation or endovascular occlusion of the feeding vessel, often an external carotid artery, is a mistake: it will not control the bleeding (3,4) because collateral vessels are rapidly recruited from either the contralateral external carotid artery or the vertebral system, as illustrated in case 2. Additionally, embolization will be difficult, if not impossible.

AVM of the mandible is effectively treated by percutaneous and/or endovascular methods without the need for surgery, as shown in these three cases. Intraarterial embolization alone is often ineffective because the arterial pedicles occlude before complete filling of the venous pouch, leading to a high recurrence rate (2).

The main embolic agent used in our cases was cyanoacrylate (ie, glue). We used a higher concentration of glue/lipiodol when injecting through a needle versus through a catheter for quicker polymerization and because, even when glued, a needle can always be retrieved safely. The volume of glue needed to cure the AVM is impossible to determine in advance. The injections should be continued until no more angiographic shunt or bleeding are observed. Treatment with cyanoacrylate has been described before with good results, but any liquid adhesive will also do (5–8).

Other nonliquid embolization materials have also been used with success (6,7,9). Chiras et al (6) described two cases treated with initial intraarterial particle embolization followed by direct puncture into the venous pouches for injection of Gelfoam (Pharmacia & Upjohn, Kalamazoo, MI) soaked in a thrombotic agent. In one of their cases, direct percutaneous deployment of a detachable balloon was also performed. Kiyosue et al (7) and Beek (8) each described case reports of successful treatment with use of intraarterial particle embolization plus transvenous coil embolization.

Reocclusion of the lytic lesion after successful treatment is expected on long term follow-up, as shown by Shira (5), Chiras (6), and Flandroy (8).

Important points can be made from our experiences treating AVM of the mandible. Careful review of the literature (3–9) and our three cases shows that AVM limited to the mandible drains almost exclusively through an intramandibular pouch. As a result, during treatment, if the pressure in this pouch is increased (ie, contrast injection in the pouch as in case 1, or occlusion of the venous exit of the pouch before complete thrombosis as in cases 2 and 3), massive oral bleeding can occur. Therefore, the procedure should be performed under general anesthesia with transnasal endotracheal intubation to allow easier access to the oral cavity. A syringe with the glue mixture should be readily available for percutaneous injection directly into the bleeding site.

Although clinically and angiographically impressive, AVMs of the mandible are easier to cure than most other facial AVMs (10) because, when occlusion of the venous pouch without bleeding to the oral cavity is achieved, the malformation is cured. This is true regardless of the type of treatment used (endovascular vs direct puncture) or the material used (glue, Gelfoam, coil, balloons, Ethibloc [Johnson & Johnson International, Brussels, Belgium]) to occlude the outlet of the AVM.

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References