Embolization of a Bleeding Maxillary Arteriovenous Malformation via the Superficial Temporal Artery after External Carotid Artery Ligation

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We report a new approach of embolization in a 15-year-old boy that presented with a massive hemorrhage from a maxillary arteriovenous malformation. Re-bleeding occurred after emergent ligation of the external carotid artery. The bleeding was successfully controlled by embolization via the superficial temporal artery.

Index terms:

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An arteriovenous malformation (AVM) of the jaw is an uncommon lesion found mainly in children. It can present with massive oral bleeding, resulting in death (1, 2). The external carotid artery (ECA) is often the feeding artery and can be ligated to control the hemorrhage. As a result, transarterial embolization is difficult or even impossible to perform when re-bleeding occurs (1, 3). We report a new approach of a successful embolization of a bleeding maxillary AVM via the superficial temporal artery (STA) after a previous ECA ligation. This technique has not yet been reported for endovascular management of a bleeding maxillary AVM.

CASE REPORT

A 15-year-old boy with a left maxillary AVM was referred to our hospital. The massive oral bleeding was previously controlled after emergent surgical ligation of the left ECA two weeks prior to admission. A contrast-enhanced computed tomography scan on admission showed a vascular lesion in the left maxilla. Elective surgical resection of the AVM was planned. While waiting for the operation, massive oral bleeding occurred. The patient was stabilized with a blood transfusion and was transferred to the interventional radiology department. Transfemoral angiography showed an interruption of the left ECA beyond the origin of the superior thyroid artery (Fig. 1A). Angiography of the bilateral internal carotid arteries (ICA) and vertebral basilar arteries (VA) revealed no blood supply to the AVM. Selective right ECA angiography demonstrated faint opacification of the distal left ECA through the small collateral arteries. The left internal maxillary artery (IMA) supplied the AVM. The left STA was slightly visualized (Fig. 1B). No attempt was made to embolize the collateral arteries because of the inability to navigate the catheter through these small vessels. Although the left STA was not palpable, trans-STA embolization was attempted. After skin preparation and sterilization, a 4-cm-long incision was made in the left temporal scalp just above the zygomatic arch. The superficial temporal artery and nerve were exposed and separated. An oblique incision was made in the STA. Using the technique described by Li et al. in rats (4), a guide-wire (0.035", Terumo,

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Tokyo, Japan) followed by a 5 Fr Cobra catheter (Terumo) was maneuvered in a retrograde fashion into the STA (Fig. 1C). After the guide-wire was maneuvered through the hairpin turn with minor resistance, the catheter was then successfully advanced into the ECA just above the ligation. Contrast injection through the catheter confirmed the IMA

to be the primary feeding artery of the AVM. The catheter was advanced super-selectively into the distal IMA. Another contrast injection demonstrated a high-flow AVM with large draining veins (Fig. 1D). No ECA-to-ICA collaterals were seen. Embolization was performed using polyvinyl alcohol particles (PVA, 350–550 μ m, Cook,

Fig. 1. Embolization of bleeding maxillary arteriovenous malformation in 15-year-old boy.

A. Selective left common carotid arteriogram shows interruption of left external carotid artery (arrow) just beyond origin of superior thyroidal artery (arrowhead).

B. Selective right external carotid arteriogram demonstrates faint opacification of distal left external carotid artery (long arrow) through multiple small collateral arteries (arrowheads). Left internal maxillary artery (open arrow) supplies arteriovenous malformation (curved arrow). Left superficial temporal artery is slightly visualized (short arrow).

C. After dissecting out superficial temporal nerve (arrowhead) and artery (arrow), 5 Fr Cobra catheter is inserted retrograde into exposed and separated left superficial temporal artery.

D. Superselective left internal maxillary arteriogram demonstrates high-flow arteriovenous malformation with large draining veins (arrowheads).

E. Post-embolization angiogram shows complete de-vascularization of arteriovenous malformation.

Bloomington, IN). A post-embolization angiogram showed complete de-vascularization of the AVM (Fig. 1E). The catheter was withdrawn and the STA was ligated at both ends of the incision. The skin incision was sutured. The duration of the procedure was less than 50 minutes and the hemorrhage was completely stopped. After the embolization, the patient complained of mild pain in the incision that was controlled with an oral analgesic. Surgical resection of the AVM was performed uneventfully five days after the embolization, and the patient was discharged seven days later without any complications. A follow-up angiogram six months later showed complete removal of the AVM. The scalp incision was completely healed with a small patch of new hair growth.

DISCUSSION

Arteriovenous malformations of the dental arcade are rare and can be fatal when massive oral bleeding occurs (1-3). Ligation of the feeding artery on an emergency basis was performed in the past to stop the hemorrhage. Because continued or recurrent bleeding can occur when the collateral vessels are recruited from either the contralateral ECA or vertebral artery, ligation of the feeding artery is no longer recommended (1, 3, 5). With the advent of endovascular treatment techniques and the use of new embolic agents, transarterial embolization is the treatment of choice for these bleeding AVMs. Because routine vascular access is closed when the ECA is ligated, transarterial embolization is difficult to perform (1, 3). Embolization after surgical exposure and puncture of the distal part of the ligated ECA has been reported (6). However, this technique is complicated and time-consuming because of the deep location of the distal ECA and the fibrosis induced by the previous surgical ligation. Gobin et al. have reported their experience of percutaneous puncture of the ECA or one of its branches after ECA ligation, including STA puncture in three cases, but the puncture-related complication rate was considerably high (7). Alternative routes include trans-venous embolization by direct trans-osseous venous puncture or by trans-femoral catheterization for delivery of various embolic agents, most often glue and coils (1-3). However, embolization by direct puncture is more invasive. More than one puncture is often needed because of the inability to predict the amount of embolic agents required. When glue is used as the embolic material in trans-venous embolization, the catheter/needle may adhere to the vessel wall. Distal migration of the glue into the pulmonary circulation may also occur. Embolization using coils is expensive and time-consuming. Most important of all,

primary trans-venous embolization carries the risk of a sudden increase of intravenous pressure with subsequent rupture and bleeding if the occlusion remains incomplete. Therefore, reduction of the arterial inflow by trans-arterial embolization at the beginning of the procedure is strongly recommended (2, 3).

With the ECA is ligated, trans-arterial embolization can be performed safely using the above-described trans-STA approach. It is necessary to perform bilateral carotid and vertebral angiography in advance to have a thorough understanding of the architecture of the AVM. Because the variation of origin and course of the STA is uncommon (8), the trans-STA approach can be successfully performed even when the artery is not palpable. As the mean diameter of the STA at the zygomatic arch is 2.73 ± 0.51 mm, a 6 Fr catheter can also be used. The feeding arteries, such as the internal maxillary and facial arteries, often branch off at sharp angles from the ECA. Therefore, the catheter should have an angled tip allowing its easy entrance into these arteries. A micro-guidewire and micro-catheter set can be used if necessary. As surgical resection was planned, we ligated the STA when the embolization was completed. In selected patients that may require a repeat embolization, the incision can be repaired to maintain patency of the STA.

A possible major complication of our technique is necrosis from embolization too distally or of too many contiguous branches. Stroke or blindness from inadvertent embolization of the ICA or VA system may result from the potential ECA-to-ICA or ECA-to-VA collaterals. Superselective embolization with large particles such as PVA particles measuring 500–750 μm in size or proximal agent may minimize complications. In our patient, superselective embolization of the distal IMA was performed using PVA particles 350–550 μm in size without any complication. Angiography for proposed embolotherapy of the ECA must include views of the extracranial and intracranial ICA and the ECA, as well as superselective views of each ECA branch before embolotherapy. Prior to embolizing posteriorly, the integrity of the VA system must also be assessed to prevent inadvertent embolization via the muscular collaterals (9). One minor complication is injury to the superficial temporal nerve. Recognition of the nerve and careful separation it from the STA may prevent this complication. A catheter-induced vasospasm can be encountered when the catheter is passed through the hairpin turn of the STA and during superselective catheterization. Very gentle manipulation with a micro-guidewire and micro-catheter set may help prevent vasospasm. When a vasospasm occurs, it can be reduced by withdrawing the catheter slightly for a period of watchful waiting or by

using vasodilators such as 100 mg of an intraarterial injection of papaverine (10).

This approach of embolization of a maxillary AVM via the STA is minimally invasive, repeatable and has little effect on the physical appearance of the patient. It may be performed on patients with hypervascular craniofacial lesions whose feeding arteries have been ligated or are too tortuous to navigate a catheter. It can also be performed urgently during active bleeding, pre-operatively and before trans-venous embolization.

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