Onyx Distal Embolization in Transarterial Embolization of Dural Arteriovenous Fistula with Subtotally Isolated Transverse-Sigmoid Sinus

A Case Report

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Summary

We describe a 39-year-old woman with dural arteriovenous fistula in the region of transverse-sigmoid sinus. A combination approach was designed to treat the fistula but the balloon failed to pass the severely stenosed proximal end of a subtotally isolated sinus. Although successfully occlusion of most feeding arteries from a single arterial injection was achieved, Onyx was found to shift to the outflow tract of the right heart ventricle and then to the lung latterly on six month follow-up.

Introduction

Advances in endovascular technology have expanded the scope of intracranial vascular fistula that is safely treated with interventional techniques. However, a dural arteriovenous fistula (DAVF) with an isolated or subtotally isolated venous sinus is a singular entity in the sense that venous drainage is through a thrombosed dural sinus, frequently accompanied by a cortical venous reflux making therapeutic disposal a challenge to neuroradiologists^{1,2}. As in our patient, the lack of endovascular access made the transarterial approach an alternative choice.

Many embolic agents have been used to treat DAVF via transarterial access. These include

microspheres of various types, silk thread, polyvinyl alcohol, avitene, and liquid agents including IBCA, NBCA and Onyx. Most complications of these treatments relate to the central nervous system; complications involving pulmonary organ systems are exceptional and rarely reported.

To the best of our knowledge, this is first time the liquid embolization agent's distal embolization has been dynamically displayed, and it is the first case Onyx was found translocated to the pulmonary system in DAVF embolization.

Case Report

A 39-year-old woman presented with left pulsatile tinnitus, dizziness and intermittent headache. She also reported right-sided limb numbness and recent hearing loss. After courses of medication without significant improvement, the patient was then transferred to the neurology service. Physical and neurological examinations were unremarkable except for left pulsatile tinnitus. Conventional digital subtraction angiography (Figure 1A,B) confirmed a DAVF draining into a subtotally isolated transverse sinus and supplied by the transosseous branches of the left occipital artery, the petrosquamous branch of the left middle meningeal artery, the branch of the accessory meningeal artery, the meningohypophyseal

trunk of the internal carotid artery, the left anterior inferior cerebellar artery, the right superior cerebellar artery and the artery of the falx cerebelli arising from the vertebral artery. The left transverse-sigmoid sinus was occluded at the distal end with severe stenosis at the proximal end where the sigmoid sinus joined the internal jugular vein. Retrograde venous drainage occurred via the left cortical vein and the left Labbé vein.

A combination of transarterial and transvenous approach was designed. Assisted by the guide wire (mirage MTI, USA), the microcatheter (marathon EV3, Irvine CA, USA) was successfully placed in the petrosquamous branch of the middle meningeal artery, adjacent to the point of fistulization. However, the balloon system (hyperform EV3, USA) failed to deliver through the venous system to the transverse-sigmoid sinus because of its proximal severe stenosis. About 0.5 ml Onyx-18 (ethylene-vinyl alcohol copolymer; ev3 Inc., Plymouth, MN, USA) was injected in 20 seconds (including catheter-filling 0.26 ml and rapid injection of 0.15 ml) to cast the DAVF but was not depicted on the angiogram. Immediate chest roentgenograms (Figure 2A) showed Onyx-18 was translocated to the outflow tract of the right heart ventricle. Heart rate was unchanged and systolic blood pressure was elevated from 120 mmHg to 160 mmHg 14 minutes later and gradually back to 120 mmHg in half an hour. The internal jugular vein was compressed to increase the pressure of the transverse sinus. Then Onyx-18 was injected again, this time it cast well and successfully occluded most of the feeding arteries (Figure 1C,D). What is more, the retrograde venous drainages disappeared on angiography.

The transosseous branches of the left occipital artery were left for second surgery. On emerging from anesthesia, the patient was symptom-free and neurologically normal. Six hours later, she experienced chest distress. She was given warfarin 1.5 mg per day intravenously, and an emergency sonogram of the heart (Figure 2B) demonstrated sliver-like Onyx trapped in the net wall of the outflow tract of the right heart ventricle. The following morning the chest distress had resolved. She was discharged seven days later, neurologically intact, on a monitoring dose of warfarin and aspirin. The sliver-like Onyx had translocated to the right lung (Figure 3) without symptoms at her six month follow-up.

Discussion

Pulmonary complications following Onyx therapeutic embolization of neurovascular lesion are rare. Murugesen et al³ reported an ARDS in a patient possibly attributed to the excretion of the solvent DMSO via the lungs. Perihilar oedema as a result of embolization of Onyx into the pulmonary artery was reported by Herman et al⁴ following the use of Onyx for the treatment of a cerebral AVM.

However, embolization of cerebral vascular lesions using other materials seem quite different. Kjellin et al⁵ reported therapeutic embolization of cerebral arteriovenous malformations in children. The chest radiographs in 35% patients showed pulmonary deposits of embolic material: cyanoacrylate in ten patients and platinum coils in two. Two of the patients with scattered cyanoacrylate deposits in the lungs developed respiratory distress that required endotracheal intubation. Pelz et al⁶ reported three patients had pulmonary symptoms within 48 hours of glue injection. One patient was embolized with an isobutyl-2-cyanoacrylate (IBCA) /pantopaque/acetic acid mixture; severe pleuritic chest pain developed two days later. The other two patients were embolized with n-butyl-2-cyanoacrylate (NBCA)/lipiodol mixtures; a cough, pleuritic chest pain and bloody sputum developed in both within 24 hours. Two patients experienced a significant drop in PO₂. All patients demonstrated significant changes (multiple peripheral radiodensities) on chest x-ray and CT chest examinations. All were treated conservatively and recovered spontaneously. The author emphasized the techniques used like flow-arrest. Fernandez et al⁷ reported a case of cyanoacrylate pulmonary embolism after embolization of a cerebral arteriovenous malformation. The patients gradually improved after one to ten days with conservative treatment. Kline et al⁸ reported symptomatic pulmonary emboli and infarction following an uneventful embolization using polyvinyl alcohol particles. Nonselective pulmonary angiogram revealed distal nonopacification, consistent with microembolization of peripheral small vessels.

Onyx seemed the most suitable agent in our patient. As a liquid agent it can be injected through the smallest most flexible catheters, which can achieve the most distal position, ideally in the point of the fistula. Coils and ca-

Figure 1 A 39-year-old woman presenting left pulsatile tinnitus, dizziness and intermittent headache proved to have a dural arteriovenous fistula with subtotally isolated transverse-sigmoid sinus. A) Right external carotid angiogram, lateral view, shows the DAVF supplied by the transosseous branches of the left occipital artery, the petrosquamous branch of the left middle meningeal artery, the branch of the accessory meningeal artery, the meningohypophyseal trunk of the internal carotid artery. B) Right vertebral angiogram, lateral view, shows the DAVF supplied by the left anterior inferior cerebellar artery, the right superior cerebellar artery and the artery of the falx cerebelli arising from the vertebral artery. Note the left transverse-sigmoid sinus occluded at the distal end and severe stenosis at the proximal end. C,D) Postoperation right external carotid angiogram, lateral view and right vertebral angiogram, anteroposterior view, show besides the transosseous branches of the left occipital artery left for latter staged surgery, all of the feeding arteries are embolized. Retrograde venous drainage via the left cortical vein and the left Labbé vein is unopacified.

Figure 2 A) Immediate chest roentgenograms show Onyx-18 is translocated to the outflow tract of the right heart ventricle. B) Emergency sonography of the heart demonstrates sliver-like Onyx trapped itself in the net wall of the outflow tract of the right heart ventricle.

theters compatible with particulate agents may not go as far distally. Deposition of the embolic material within the point of fistulas is essential for cure of the lesion in one treatment session. Proximal occlusion of branches will result in recanalization by adjacent branches and persistence of the lesion.

Another reason is that Onyx is a biocompatible polymer containing ethyl-vinyl alcohol dissolved in dimethyl sulfoxide (DMSO). It is characterized by a different solidification

process consisting of a copolymer precipitation. Unlike NBCA or IBCA polymerization, which is triggered by contact with ionic solutions such as blood or saline, Onyx presents a liquid form when injected and a solid form when in contact with blood through precipitation after the dimethyl sulfoxide evaporates, thus allowing for slower and longer injection rates, which can be better controlled. This should theoretically result in a more effective filling of the DAVF shunt, thus reducing the

Figure 3 The chest radiography preoperation, postoperation and at six month follow-up. Note the sliver-like Onyx translocated to the right lung.

risk of the microcatheter adhering to the endothelium, or even of its untimely obstruction. Because Onyx is not absorbable, it is capable of producing permanent vascular occlusion. What is most important, Onyx has a lava-like flow pattern within blood vessels without any fragmentation during injection^{9,10}. Even on the rare occasions it has penetrated the venous wall and translocated to the heart and lungs, Onyx would not scatter everywhere like NBCA, IBCA and other particle agents⁵⁻⁸, resulting in a diffuse pulmonary lesion.

However, even with the optimal cure of recent cases and the safety of the Onyx reported in the literature, the complication of the Onyx in transarterial embolization of DAVF cannot be overlooked, especially its distal embolization. Several factors were thought to be involved in Onyx solidifying at the fistulization point: the differential pressure near the fistula, the blood fluid direction, the speed of the blood fluid, the location of the fistulization point, the strength of the surgeon injecting the Onyx, etc. Our experience of 38 high-flow DAVFs demonstrated distal embolization of Onyx in only two patients (5%) (REF).

In these cases the strength of injection was thought to be one of the factors for successful embolization: sudden injection of a bundle of Onyx would bounce against the vessel wall and into the network of feeding arteries. The balloon we used was aimed at increasing the sinus pressure temporarily and totally curing in one session, even if there was a risk of redirecting the Onyx into the cortical vein. The reason might be that in this type of fistula the cortical vein was thought to result from the increase in sinus pressure and directly connect with the sinus. As some other factors are undetectable in clinical practice, great attention should be paid to Onyx distal embolization, as well as brisk management.

If the Onyx translocated to heart, the threat of thrombus formation in the heart and possible embolization of the lung is very real. Besides full heparinization during the procedure, it is our practice to treat our patients who undergo Onyx distal embolization with 1.5 mg of warfarin per day to maintain the INR within the range of 2.0-3.0 after the procedure. Emergency sonography of the heart is essential to evaluate the position, shape and the anterior

and posterior pressure of the foreign substance. Furthermore, to lessen the incidence of delayed thromboembolism and long-term possible pulmonary embolization and infarction, patients are maintained on warfarin for a total of three to six months in addition to two years of 100mg aspirin per day. Use of warfarin increases the risk of bleeding, both internally and externally. To minimize the risk, people taking warfarin must have periodic blood tests to see how much their blood is anticoagulated. Doctors then use the blood test result to adjust the dose of warfarin.

Surgery may be needed for patients with persistent shortness of breath or pulmonary hypertension. The likelihood of dying from pulmonary embolism is very low, but massive pulmonary embolism can cause sudden death. Most deaths occur before the diagnosis is made, often within one to two hours of the embolism occurring. If a person is alive when diagnosed, the chance of survival is about 95%¹¹. Important factors include the size of the embolus, the size and number of pulmonary arteries blocked, and the person's overall health status. A person with normal heart and lung function usually survives unless the embolus blocks half or more of the pulmonary arteries.

Conclusions

This solitary case merits scrutiny in the long-term clinical follow-up, and we anticipate further limitations of this technique will be discovered with its use in future patients. Transarterial Onyx embolization can clinically and anatomic obliterate DAVF with subtotal isolated sinus. What is more, embolization allowed occlusion of multiple arterial feeding arteries from a single arterial injection.

Technically, the embolization was optimized when a microcatheter position immediately adjacent to the point(s) of fistulization was achieved, but great attention should be paid to Onyx distal embolization. Considering the high number of embolization procedures done nowadays, this complication must be borne in mind. Although not supported by the data presented above, embolization may be a life-saving procedure in many patients with vascular malformations and will outweigh the risk of pulmonary embolism.

References

- 1 Houdart E, Saint-Maurice JP et Al: Transcranial approach for venous embolization of dural arteriovenous fistulas. *J Neurosurg* 97: 280-286, 2002.
- 2 Gobin YP, Houdart E et Al: Percutaneous transvenous embolization through the thrombosed sinus in transverse sinus dural fistula. *Am J Neuroradiol* 14: 1102-1105, 1993.
- 3 Murugesan C, Saravanan S et Al: Severe pulmonary oedema following therapeutic embolization with Onyx for cerebral arteriovenous malformation. *Neuroradiology* 50: 439-442, 2008.
- 4 Herman TE, Siegel MJ et Al: Cerebral arteriovenous fistula to pulmonary artery onyx embolization. *J Perinatol* 27: 238-240, 2007.
- 5 Kjellin IB, Boechat MI et Al: Pulmonary emboli following therapeutic embolization of cerebral arteriovenous malformations in children. *Pediatr Radiol* 30: 279-283, 2000.
- 6 Pelz DM, Lownie SP et Al: Symptomatic pulmonary complications from liquid acrylate embolization of brain arteriovenous malformations. *Am J Neuroradiol* 16: 19-26, 1995.
- 7 Fernandez P, Loayza P et Al: Cyanoacrylate pulmonary embolism after embolization of a cerebral arteriovenous malformation. Report of one case. *Rev Med Chil* 132: 489-492, 2004.
- 8 Kline JN, Ryals TJ et Al: Pulmonary embolization and infarction. An iatrogenic complication of transcatheter embolization of a cerebral arteriovenous malformation with polyvinyl alcohol sponge. *Chest* 103: 1293-1295, 1993.
- 9 Weber W, Kis B et Al: Endovascular treatment of intracranial arteriovenous malformations with onyx: technical aspects. *Am J Neuroradiol* 28: 371-377, 2007.
- 10 Vanninen RL, Manninen I: Onyx, a new liquid embolic material for peripheral interventions: preliminary experience in aneurysm, pseudoaneurysm, and pulmonary arteriovenous malformation embolization. *Cardiovasc Intervent Radiol* 30: 196-200, 2007.
- 11 Tuttle-Newhall JE, Rutledge R et Al: Statewide, population-based, time-series analysis of the frequency and outcome of pulmonary embolus in 318,554 trauma patients. *J Trauma* 42: 90-99, 1997.

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