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# Onyx removal after embolization of a superior sagittal sinus dural arteriovenous fistula involving scalp artery

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## Abstract

**Background:** Most dural arteriovenous fistula (DAVF) in superior sagittal sinus (SSS) requires multimodal treatment. Onyx embolization is useful for DAVF; however, scalp artery embolization has cast extrusion risk.

**Case Description:** A 59-year-old male presented with involuntary movements of both legs and progressive dementia. Cerebral angiography demonstrated the DAVF in the SSS fed by bilateral superficial temporal, occipital, and middle meningeal arteries. The posterior SSS was thrombosed, and the main drainers were cortical veins. Combined treatment with transarterial embolization using Onyx and transvenous embolization using coils was performed. Although symptoms were improved, a small DAVF remained. Two months later, Onyx cast extrusion through the scalp was observed, requiring removal and debridement because of infection at the extrusion sites. Surgery for the residual DAVF would be difficult because of scalp condition; therefore, an additional endovascular treatment was conducted, completely occluding DAVF.

**Conclusion:** Onyx embolization is useful for DAVF; however, scalp artery embolization has cast extrusion risk. Therefore, scalp infection should be considered because it may preclude additional surgical procedures.

**Key Words:** Complication, dural arteriovenous fistula, Onyx, scalp artery, superior sagittal sinus

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## INTRODUCTION

Currently, transarterial embolization (TAE) with the liquid embolic Onyx is used for the treatment of the superior sagittal sinus (SSS) dural arteriovenous fistulas (DAVFs).[1,3,15,19,23,26-28,31] We present a case of spontaneous Onyx extrusion from the scalp 2 months after embolization. Although there is one previous report of Onyx extrusion from the scalp, the patient demonstrated spontaneous wound healing.[27] To the best of our knowledge, this is the first case report to document scalp infection requiring debridement because of Onyx extrusion from the scalp. Treatment strategies are discussed based on a literature review.

## CASE HISTORY

A 59-year-old male presented with involuntary movements of both legs and progressive dementia. Magnetic



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resonance imaging and cerebral angiography revealed SSS-DAVF with occlusion of the posterior one-third of SSS. This SSS-DAVF was fed by bilateral occipital arteries (OAs), superficial temporal arteries (STAs), thin middle meningeal arteries (MMAs), and meningeal branches of the vertebral artery. Anterograde drainage was not observed through SSS, whereas drainage routes through dilated medullary and cortical veins (CV) were identified. Iodine-123-N-isopropyl-p-iodoamphetamine single-photon emission computed tomography revealed hypoperfusion of the vertex sides [Figures 1 and 2].

## First embolization

Right femoral artery puncture was performed under general anesthesia. A microcatheter (Marathon; eV3 Neurovascular, Irvine, CA, USA) was navigated to the distal left OA for Onyx-18 injection, causing incomplete DAVF obliteration. Onyx embolization was performed through the left STA, resulting in a partial reduction in blood flow from the left feeding arteries [Figure 3a].

## Second embolization

We performed Onyx embolization from the right OA. However, Onyx infiltration into the fistula point was unsuccessful; therefore, transvenous embolization (TVE) was conducted. Through the 4F guiding catheter, a microcatheter (Neurodeo 10; Medico Hirata, Tokyo, Japan) was advanced over a microguidewire (Chikai 0.014 inch; Asahi Intecc, Nagoya, Japan) into the posterior third of the thrombosed SSS. The fistulous portion of SSS was tightly packed from the anterior to posterior end with coils of various sizes. However, little cortical venous drainage to the left parietal lobe was present [Figure 3b]. Nonetheless, involuntary movement disappeared and dementia improved after the second embolization procedure.

Two months after the last Onyx embolization, the patient complained of scalp pain along the right OA. Redness and swelling of his scalp were clearly observed exactly above OA, and Onyx casts were partially exposed within lesions [Figure 4a]. *Staphylococcus aureus* and *Serratia marcescens* were detected in two separate cultures. Despite a 1-week treatment with systemic antibacterial agents, the subcutaneous exudate increased necessitating debridement to treat the scalp infection. A scalp incision was created exactly above the right OA. This revealed naked Onyx casts without surrounding arterial structures. Although granulation tissue was observed around the Onyx casts, no abscesses were observed [Figure 4b]. The Onyx casts were totally excised. No bacteria were detected, but necrosis associated with prominent inflammatory histiocyte and neutrophil infiltration was revealed histologically. The incision site completely healed after debridement.

## Third embolization

Follow-up angiography revealed left MMA growth. Onyx embolization through the left MMA was performed, and complete occlusion of the fistula was achieved [Figure 3c]. The postoperative course was uneventful. He was discharged home, and he subsequently returned to

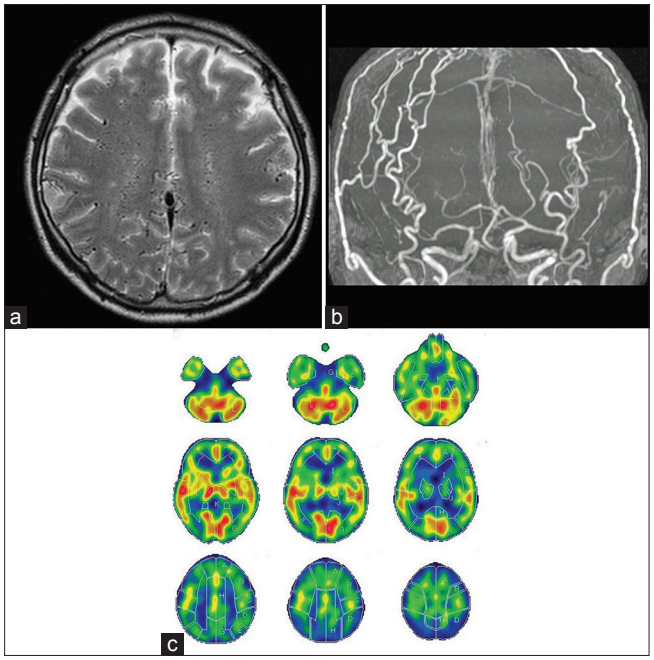


**Figure 1: (a) Preoperative T2-weighted magnetic resonance image revealing multiple aberrant medullary veins. (b) Magnetic resonance angiography revealing expanded bilateral occipital arteries and superficial. (c) Iodine-123-N-isopropyl-p-iodoamphetamine single-photon emission computed tomography revealing hypoperfusion of bilateral vertex sides of the frontal, parietal, and occipital lobules**

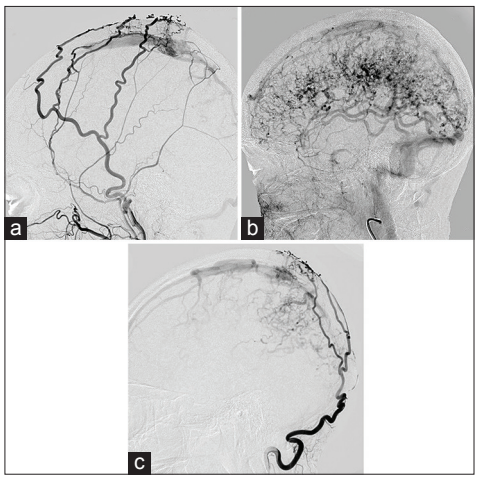


**Figure 2: Angiographic images (lateral view). (a) Preoperative right external carotid angiography showing a dural arteriovenous fistula in the superior sagittal sinus fed by the right superficial temporal artery and thin middle meningeal artery. (b) Preoperative right external carotid angiography in the venous phase showing occlusion of the posterior one-third of superior sagittal sinus. The dural arteriovenous fistula is drained via anterograde flow by cortical and medullary veins into the vein of Labbe or deep venous system and subsequently into the vein of Galen. (c) Preoperative right occipital artery angiography showing the dural arteriovenous fistula fed by occipital artery**

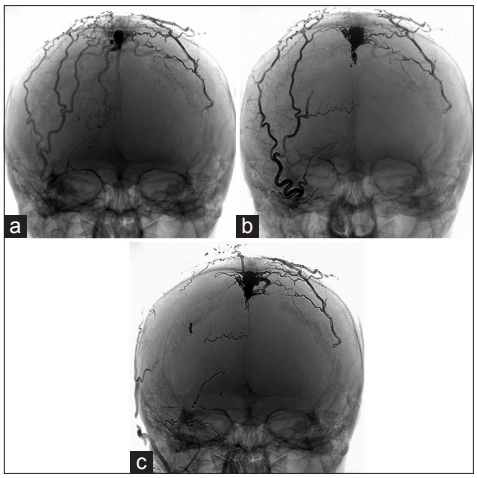


**Figure 3: Angiographic images (anteroposterior view). (a) After the first embolization, right occipital artery angiography showing a residual dural arteriovenous fistula after left occipital artery and superficial temporal artery embolization using Onyx. (b) After the second embolization, right carotid artery angiography showing a reduction of the shunt and remaining drainer. Right occipital artery is occluded by Onyx. The proximal segment of the right occipital artery is highly tortuous. Compared to the left occipital artery, the right occipital artery is occluded from the proximal segment. The fistulous portion of superior sagittal sinus is packed using coils of various sizes. (c) After the third embolization, right external carotid angiography showing complete obliteration of the dural arteriovenous fistula. Residual dural arteriovenous fistula is occluded by Onyx. The Onyx cast in the right occipital artery was extruded**

work. Follow-up angiography 6 months later showed no recurrence of DAVF.

## DISCUSSION

SSS-DAVFs account for approximately 8% of all DAVFs,[6] and the reported cases have used various treatments. To the best of our knowledge, 27 cases with sufficient hemodynamic description for definitive diagnosis of SSS-DAVF involving scalp arteries have been reported [Table 1].[1-3,5,7-10,12,14-17,20,22-24,26-31] Most cases were middle-aged males. Drainage to CV was found in 25 cases (25/27, 92.6%), indicating that SSS-DAVF is the most aggressive form of DAVF. Various SSS-DAVF treatment options involving scalp arteries have been reported including TAE, TVE, surgical TAE or TVE, surgery, radiosurgery, and various combinations.

TAE alone was performed in seven cases (7/27, 25.9%). TAE with Onyx for DAVF has been increasingly used since Onyx was first available in the year 2000.[1,3,15,19,23,26-28,31] Nine cases of SSS-DAVF treated with Onyx have been reported [Table 2]. Because of its nonadhesive nature and penetration characteristics, TAE with Onyx may be effective and safe if MMA is the only or main feeding artery. However, complete DAVF embolization with multiple feeding arteries including scalp arteries is difficult using Onyx alone. Only two

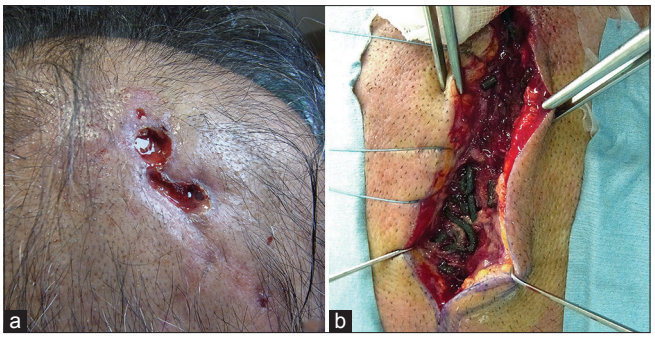


**Figure 4: (a) A photograph showing regions of scalp erosion and swelling above occipital artery. (b) An intraoperative photograph showing naked Onyx casts**

cases were successfully treated using a simple TAE with Onyx; most cases required additional treatment.

Combined treatment was the single most common choice, performed in 14 cases (51.9%). Total DAVF obliteration involving SSS is difficult because of their midline location and presence of multiple bilateral feeding arteries, sinus occlusion/stenosis, and scalp artery supply and critical cortical drainage pathway involvement. Therefore, simple TAE or TVE is usually not sufficient, similar to surgery alone because of the danger of intraoperative bleeding or the requirement of a large craniotomy to expose all drainage routes. Therefore, a combined treatment is indicated in most cases.[2,6,9-11,13,18,25]

In our case, TAE with Onyx was selected first. Because the bilateral MMAs were very thin to insert a microcatheter, we performed TAE from the left STA and OA. However, this only reduced blood flow through the fistula. The cause was high tortuosity of OA and STA. When injected from the proximal OA, Onyx only occluded OA itself and did not reach the fistula. Although an improvement of symptoms was noted after additional TVE, complete embolization was not achieved. We considered that surgical treatment would be useful as further treatment because the residual DAVF was quite localized. However, surgery would have been difficult because of scalp infection/necrosis. Consequently, endovascular therapy was considered a better treatment.

Onyx (eV3 Neurovascular, Irvine, California, USA), an ethylene vinyl alcohol copolymer dissolved in dimethyl sulfoxide (DMSO), is increasingly used as an embolic agent in intracranial vascular malformation management. Transarterial Onyx embolization is now an established DAVF treatment. Murayama *et al.* found that Onyx could cause inflammatory changes in the subacute and chronic phases after embolization.[21] They found that rapid intra-arterial DMSO injection caused angiographic vasospasm and histological endothelial necrosis, whereas slow DMSO injection evoked minimal or no angiographic vasospasm and adventitial inflammation.[4]

**Table 1: Summary of superior sagittal sinus dural arteriovenous fistula involving scalp arteries in the literature**

| Author (year) | Age/sex | Feeder | Drainer | Sinus patency | Treatment | Outcome |
|---|---|---|---|---|---|---|
| Halbach (1986) | ND | MMA, OA, STA | SSS | ND | Surgery | ND |
| | ND | MMA, STA, OA, MCA, VA | SSS | ND | Surgery | ND |
| | ND | MMA, STA | SSS, CV | ND | Combined | ND |
| Kurl (1996) | 46/male | OA, MMA | SSS, CV | Patient | TAE | GO |
| Cloft (1997) | 74/male | MMA, STA | SSS, CV | SSS, OC | TVE | GO |
| Kawaguchi (2000) | 62/male | MMA, STA, OA | SSS, CV | Patient | TAE | GO |
| Bertalanffy (2001) | 51/male | OA, PAA, STA, MMA, ICA | SSS, CV | SSS, OC | TAE | GO |
| | 57/female | MMA, STA | CV | Patient | Combined | GO |
| | 71/male | Falx A, STA, OA | CV | SSS, OC | Combined | GO |
| Fukai (2001) | 38/female | Scalp A, MMA, ACA, STA | SSS, CV | Patient | TAE | GO |
| Houdart (2002) | 21/male | OA, VA | SSS, CV | SSS, OC | Combined | GO |
| Nishio (2002) | 57/female | STA, MMA, OA, falx A | CV | Right TS, OC | Combined | GO |
| Arat (2006) | 54/male | ECA, APA, ACA, MCA | SSS, CV | Patient | TAE* | GO |
| Kong (2007) | 53/male | OA, VA, MMA | CV | Torcular OC, TS, OC | Combined | GO |
| Yoshioka (2007) | 51/male | STA, MMA, OA | CV | Patient | TAE | GO |
| Toyota (2008) | 68/male | STA, MMA, OA | CV | SSS, OC | TVE | GO |
| Chai (2011) | 45/male | MMA, OA | CV | ND | TAE* | GO |
| Mitsuhara (2011) | 70/male | STA, MMA, OA | SSS, CV | Right SS, OC | Surgery | ND |
| Ohara (2012) | 61/male | MMA, STA, OA | CV | SSS, OC | Combined | GO |
| Spiotta (2012) | 68/male | STA, OA, MMA | SSS, CV | SSS, ST | Combined* | Dead |
| Kim (2013) | 77/male | MMA, STA, ACA, VA | LV, SPS | SSS, OC | Combined* | No change |
| Fujii (2014) | 69/male | STA, OA | CV | SSS, OC | TVE | GO |
| Shimizu (2014) | 75/male | MMA, OA, STA, AFA | CV | Bilateral TS, OC | Combined* | GO |
| Imazeki (2015) | 72/male | STA, MMA, OA | CV | Patient | Combined | GO |
| Singla (2015) | 60/male | OA, MMA, Falx A | CV | ND | Combined* | GO |
| Oh (2015) | 66/female | MMA, STA, OA | CV | SSS, OC | Combined* | GO |
| Zhang (2015) | 23/male | MMA, STA, OA, MHT, PCA | CV | Right TS, OC, left TS, ST | Combined* | GO |
| Our case (2015) | 59/male | STA, OA, MMA, VA | SSS, CV | SSS, OC | Combined* | GO |

*Treated with Onyx. AFA: Anterior falx artery, APA: Ascending pharyngeal artery, CV: Cortical veins, ECA: External carotid artery, MMA: Middle meningeal artery, OA: Occipital artery, OC: Occlusion, SSS: Superior sagittal sinus, SPS: Superior parietal sinus, ST: Stenosis, STA: Superficial temporal artery, TAE: Transarterial embolization, TS: Transverse sinus, TVE: Transvenous embolization, LV: Vein of the Labbe, ND: No description, MHT: Meningohypophyseal trunk, PCA: Posterior cerebral artery, A: Vertebral artery, MCA: Middle cerebral artery, PAA: Posterior auricular artery, ICA: Internal carotid artery, ACA: Anterior cerebral artery, GO: Good outcome

**Table 2: Summary of superior sagittal sinus dural arteriovenous fistula treated with Onyx in the literature**

| Author (year) | First treatment | Second treatment | Third treatment |
|---|---|---|---|
| Arat (2006) | TAE from MMA | - | - |
| Chai (2011) | TAE from MMA | - | - |
| Spiotta (2012) | TVE from SSS with balloon assist, stent angioplasty of SSS | - | - |
| Kim (2013) | TVE with coil | TAE from MMA | - |
| Shimizu (2014) | TAE from MMA | TAE from STA | TAE from OA with direct puncture |
| Singla (2015) | TAE from OA | Surgical obliteration | - |
| Oh (2015) | TAE from MMA and STA | TAE from MMA with transcranial direct puncture | - |
| Zhang (2015) | TAE from OA, stent angioplasty of TS | TAE from MMA with balloon assist, TVE with coil | TAE from MMA with balloon assist |
| Our case (2015) | TAE from OA and STA | TAE from OA, TVE with coil | TAE from MMA |

MMA: Middle meningeal artery, OA: Occipital artery, SSS: Superior sagittal sinus, STA: Superficial temporal artery, TAE: Transarterial embolization, TS: Transverse sinus, TVE: Transvenous embolization

Based on a previous report,[27] we assume that the spontaneous Onyx extrusion was secondary to inflammation. A combination of inflammation, ischemia, and radiation damage could increase extrusion and scalp infection risks. A larger amount of injected Onyx may induce more widespread scalp inflammation, whereas embolization from the proximal segment of the scalp arteries may induce scalp ischemia. Embolization of a

long segment, which requires a longer treatment time, would increase the risk of a radiation-induced scalp disorder. Here, blood vessel-like structures were not observed around the extruded Onyx casts intraoperatively. Vascular structures were replaced by granulation tissue, suggesting chronic inflammation caused by Onyx. We suggest that if embolization through the tortuous scalp arteries is required, surgical exposure of the scalp arteries near the fistula point and a minimal amount of Onyx may be desirable. Onyx is a foreign material; therefore, debridement is required when infection occurs.

We described a case of Onyx cast extrusion through scalp arteries and ensuing infection following embolization of OA using Onyx. Most DAVFs in SSS involving scalp arteries require multimodality treatment. Special care should be taken when embolizing scalp arteries using Onyx as these may be prone to inflammatory damage, resulting in Onyx cast extrusion and infection.

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## Conflicts of interest
There are no conflicts of interest.

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