



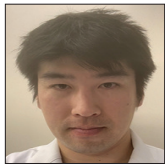
## Case Report

# Sphenoid wing dural arteriovenous fistula: A case report and literature review

Yu Shimizu, Kazuhiko Tokuda, Cheho Park

Department of Neurosurgery, Japan Organization of Occupational Health and Safety Toyama Rousai Hospital, Toyama, Japan.

E-mail: \*Yu Shimizu - bleuler3a@yahoo.co.jp; Kazuhiko Tokuda - bleuler5a@yahoo.co.jp; Cheho Park - bleuler6a@yahoo.co.jp



### \*Corresponding author:

Yu Shimizu,  
Department of Neurosurgery,  
Japan Organization of  
Occupational Health and  
Safety Toyama Rousai Hospital,  
Toyama, Japan.

[bleuler3a@yahoo.co.jp](mailto:bleuler3a@yahoo.co.jp)

Received : 24 August 2020  
Accepted : 18 November 2020  
Published : 16 December 2020

DOI  
[10.25259/SNI\\_571\\_2020](https://doi.org/10.25259/SNI_571_2020)

Quick Response Code:



## ABSTRACT

**Background:** Sphenoid wing dural arteriovenous fistula (SWDAVF) is rare that is typically fed by middle meningeal artery feeders and that drain through the sphenoparietal sinus or middle cerebral vein. Here, we report a case of SWDAVF treated by coils placed in the venous aneurysm through the contralateral cavernous sinus (CS).

**Case Description:** A 37-year-old woman was admitted to our hospital with headache and bilateral oculomotor nerve palsy. Magnetic resonance images and an angiogram showed a venous aneurysm in the right middle cranial fossa. A DAVF, consisting of two main feeders, was diagnosed based on the angiogram findings. The fistula drained into the left inferior petrosal sinus (IPS) through the left CS and right IPS. Given the remarkable extent of venous ectasia together with the headache and right abducens nerve paralysis, endovascular treatment was initiated. A transvenous approach through the right IPS was not feasible, as it is strenuous to insert the microcatheter into the right IPS. Thus, we tried an approach through the left IPS. The venous aneurysm was embolized with coils. The postoperative course was uneventful, and postoperative cerebral angiography confirmed disappearance of the fistula.

**Conclusion:** A SWDAVF is extremely rare. In our case, since the AVF drained into the contralateral CS, contralateral ocular symptoms occurred. Endovascular occlusion of the venous aneurysm and fistula was achieved through a transvenous approach.

**Keywords:** Cavernous sinus, Dural arteriovenous fistula, Endovascular, Venous aneurysm

## INTRODUCTION

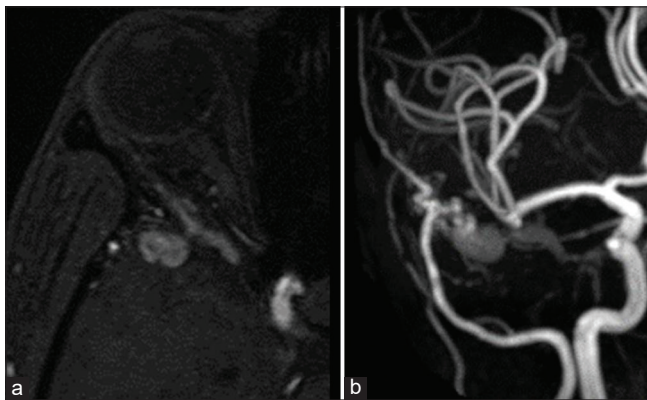
Intracranial dural arteriovenous fistula (DAVF) is sporadic, pathologic arteriovenous connections that most commonly involve the wall of a major dural venous sinus.<sup>[15]</sup> The sphenoid bone forms the foundation of the anterior and middle cranial fossae. It is a centrally placed bone, through which critical neurovascular connections are transmitted through vital foramina. DAVF of the lesser sphenoid wing is often considered to be abnormal connections between the middle meningeal artery (MMA) and the sphenoparietal sinus.<sup>[5,18]</sup> Most sphenoid wing DAVF (SWDAVF) are fed by the MMA and drain into the sphenoparietal sinus and the superficial middle cerebral vein (SMCV).<sup>[11]</sup> In our case, the DAVF drained into the cavernous sinus (CS), mimicking CS DAVF. Endovascular occlusion of fistulous connections by transarterial or transvenous approaches is the primary therapeutic strategy to cure these potentially dangerous lesions. Here, we describe a case of DAVF in the sphenoid wing accompanied with ocular symptoms; we successfully treated this patient with transvenous embolization.

This is an open-access article distributed under the terms of the Creative Commons Attribution-Non Commercial-Share Alike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as the author is credited and the new creations are licensed under the identical terms.

©2020 Published by Scientific Scholar on behalf of Surgical Neurology International

## CASE PRESENTATION

A 37-year-old woman presented with headache, bilateral ptosis, and right proptosis. She denied a history of head trauma and hypertension, and her medical history was unremarkable. Her laboratory data were all normal. Physical examination revealed bilateral oculomotor nerve palsy. Magnetic resonance (MR) imaging demonstrated varix in the right middle cranial fossa [Figure 1a and b]. Cerebral angiography revealed a high-flow, multichannel fistula between the right MMA and a venous aneurysm wall at the right greater sphenoid wing, which drained into the left CS superior orbital vein (SOV) and inferior petrosal sinus (IPS). The petrosal outlets of the right CS were occluded on the way. On the venous side, retrograde venous drainage was observed from the right SWL to the right facial vein through a right SOV with venous ectasia. Second, the venous drainage from the DMCV drained into the right CS and then the left CS, through the inter-CS (ICS), and into the left IPS [Figure 2a-c]. No connection between the SWL and the basal vein was observed. Given the remarkable ectasia of the draining vein, accompanied by the presence of headache



**Figure 1:** Time-of-flight magnetic resonance (MR) images (a) and MR angiography (b) show a venous aneurysm in the right middle cranial fossa and dural arteriovenous fistula around the aneurysm (white arrow).

and right abducens nerve paralysis, endovascular treatment was initiated.

Our approach was through the left IPS since the right IPS was occluded. A 6 Fr FUBUKI (ASAHI INTECC, Nagoya, Japan) guiding catheter was placed in the left internal jugular vein. A 4.2 Fr FUBUKI distal access catheter (ASAHI INTECC, Nagoya, Japan) was introduced through the left IPS and then placed in the right CS at the orifice of the sphenoparietal sinus. Excelsior SL-10STR (Stryker, Kalamazoo, MI, USA) and ASAHI CHIKAI (ASAHI INTECC, Nagoya, Japan) were advanced into the venous aneurysm of the SWL. Following the introduction of the microcatheter into the SWL, superselective angiography suggested a high-flow, multichannel fistula between the right petrosal branch of the MMA and a venous aneurysm wall [Figure 2c]. We assigned this area as the fistulous point, and the venous aneurysm was occluded using a Target 360 soft coil (Stryker Neurovascular) followed by the application of additional down sized coils. Complete obliteration of the venous aneurysm was achieved after the insertion of 10 coils [Figure 3a]. A final right external carotid angiography showed disappearance of the fistula [Figure 3b and c]. The postoperative course was uneventful. After the procedure, the symptoms of headache and diplopia improved.

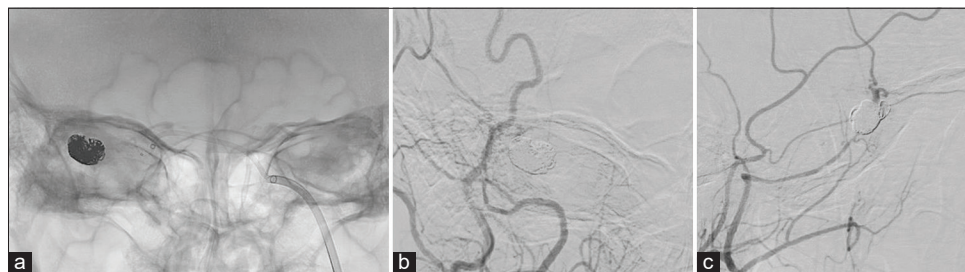
Postoperative cerebral angiography and time-of-flight MR image on the 3<sup>rd</sup> month showed disappearance of the venous aneurysm and fistula [Figure 4a and b].

## DISCUSSION

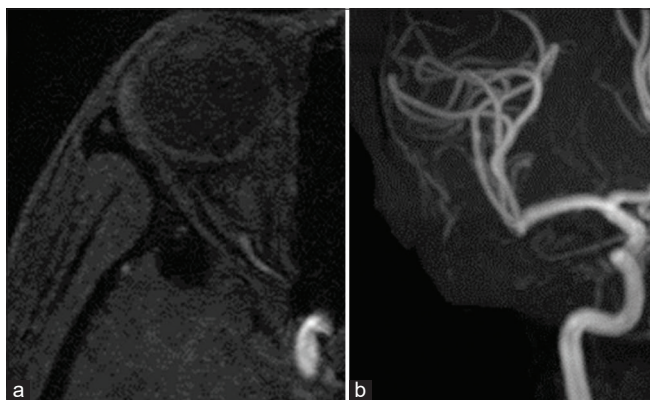
DAVF within the greater and lesser sphenoid wings has vascular features distinctive from the CS.<sup>[14]</sup> The sphenoidal group of cerebral veins formed by the terminal ends of the superficial Sylvian, and occasionally, the deep Sylvian veins drain into the sphenoparietal or CS. It less commonly drains into the sphenobasal or sphenopetrosal sinuses that course on the inner surface of the sphenoid bone.<sup>[20]</sup> The incidence rate of SWDAVF was reported to be 1.54% of all



**Figure 2:** (a and b) Right external carotid artery injection, frontal view. A large venous varix rapidly fills with early drainage into the superior orbital vein, left cavernous sinus, and inferior petrosal sinus. The feeding of the dural arteriovenous fistula (DAVF) through the middle meningeal artery is observed. (c) Superselective catheterization of the middle meningeal artery (MMA) shows feeding of the DAVF through the petrosal branch of the MMA.



**Figure 3:** (a) Venous aneurysm is completely obliterated by the coil mass. (b) Frontal view. (c) Lateral view, final right external carotid angiography showing complete obliteration of the fistula.



**Figure 4:** Time-of-flight magnetic resonance (MR) images (a) and MR angiography (b) demonstrate that the venous aneurysm and dural arteriovenous fistula disappeared after coil embolization.

intracranial DAVF, and it is suggested that a DAVF diagnosis might be difficult because it could easily be confused with a CS DAVF.<sup>[16]</sup> In our case, venous drainage from the right CS to the left CS was shown in the early venous phase. Arteriovenous fistulae cause an increase pressure of the left CS and also right CS; hence, contralateral ocular symptoms occurred. Clinical manifestations include ptosis, global amnesia, headache, visual field defects, and dizziness.<sup>[13,19]</sup> Although few patients exhibit aggressive presentation, such as intracranial hemorrhage or progressive neurological deficits, treatment is strongly recommended due to the presence of varix similar to our case.<sup>[1]</sup>

The treatment options for SWDAVF have been described in previous reports and include TAE, TVE, and surgical obliteration. However, TVE is the primary therapeutic strategy for the curative treatment of DAVF. If it is impossible to approach the affected sinus through the IPS, it is necessary to consider another venous access route, such as the facial vein or the vein of Galen.<sup>[6]</sup> In the current era of treating DAVF with curative embolization using Onyx, TAE of fistulous connections is the primary treatment technique.<sup>[21]</sup> Flow control using coils, acrylics, or temporary balloon occlusion is useful for high-flow shunts to decrease the likelihood of distal embolic migration and to improve

the penetration of embolic material into the arteriovenous connections.<sup>[16]</sup>

We summarized the clinical data of SWDAVF [Table 1].<sup>[2-4,7-10,12,16,17,19,22,23]</sup> SWDAVF is rare, and only 15 cases have been reported in the literature, including our case. The median patient age was 52.9 years (range, 27–71 years), and the patients were predominantly male (80%). Nine out of 15 patients had varix. The four cases who exhibited cerebral symptoms had venous drainage related to reflux into the SMCV or through the petrosal veins into the perimesencephalic and cerebellar veins. In these patients, however, cerebral symptoms and SAH were always concomitantly present with CS symptoms, such as proptosis and retinal hemorrhage. In addition to the SMCV, the CS and SOV were seen during angiography. Five cases presented with orbito-ocular or CS symptoms and had antegrade (through the IPS) and/or retrograde (through SOV) venous drainage routes.

Shi *et al.* reported two cases treated by successful endovascular treatment.<sup>[16]</sup> Embolization through a transarterial or transvenous approach is the primary therapeutic strategy for these lesions. Our patient was fortunately cured by only a transvenous approach.

Endovascular treatment has been favored because the draining vein just proximal to the fistula point at the lesser sphenoid wing is easy to access through the IPS and CS. Six patients underwent endovascular treatment only, four patients underwent surgery only, and five underwent both [Table 1].

Although arterial embolization for SWDAVF has only been performed to reduce the risk of the surgical approach, successful endovascular occlusion for SWDAVF has been reported recently.<sup>[13]</sup> Fukuda *et al.* reported two cases with successful endovascular treatment and concluded that SWDAVF could be cured safely by endovascular treatment with proper strategy and instruments.<sup>[3]</sup>

## CONCLUSION

Here, we report the rare patient of SWDAVF presenting with ocular symptoms. TVE was performed, and the patient had



**Table 1:** Summary of the 15 cases of venous embolization of sphenoid wing dural arteriovenous fistula.

Author/year	Age/sex	Symptoms treatment	Feeding artery	Draining vein	Varix	Treatment	Angiographic outcome
Thompson <i>et al.</i> <sup>[19]</sup>	44/M	Right ptosis	MMA	BVR, SMCV	-	TAE + Surgery	Complete
Ezura <i>et al.</i> <sup>[2]</sup>	40/M	Transient global amnesia	MMA	BVR, SMCV	+	TAE + Surgery	Complete
Nomura <i>et al.</i> <sup>[10]</sup>	59/F	Subarachnoid hemorrhage	MMA	SMCV	-	Surgery	Complete
Rezende <i>et al.</i> <sup>[13]</sup>	45/M	Headache	MMA, AMA, FRA	BVR, SMCV	+	TAE	Complete
Zhou <i>et al.</i> <sup>[23]</sup>	42/M	Subarachnoid hemorrhage	MMA	SMCV	+	TAE + Surgery	Complete
Tanaka <i>et al.</i> <sup>[17]</sup>	61/M	Hemianopsia	MMA	BVR, SMCV	+	Surgery	Complete
Shi <i>et al.</i> <sup>[16]</sup>	62/M	Hemianopsia	MMA, AMA, FRA	BVR, SMCV	+	TAE+TVE	Complete
Fukuda <i>et al.</i> <sup>[3]</sup>	58/M	Dizziness	MMA, AMA, FRA	BVR, SMCV	+	TAE+TVE	Complete
Watanabe <i>et al.</i> <sup>[22]</sup>	69/M	-	MMA	BVR, SMCV	+	Surgery	Complete
Murakami <i>et al.</i> <sup>[8]</sup>	68/M	-	AMA, MMA, AFR	BVR	+	TAE	Complete
Nakajima <i>et al.</i> <sup>[9]</sup>	71/M	Pontine hemorrhage	MMA	BVR	-	Surgery	Complete
Misaki <i>et al.</i> <sup>[7]</sup>	42/F	Right ptosis, chemosis	MMA	SOV	-	TVE	Complete
Park and Lee. <sup>[12]</sup>	69/M	Subarachnoid hemorrhage	MMA	SOV	-	TAE	Complete
Kandyba <i>et al.</i> <sup>[4]</sup>	27/M	Headache	MMA	SMCV	-	TAE	Complete
Present case	37/F	Bilateral ptosis, right proptosis, headache	MMA	Contralateral CS, SOV	+	TVE	Complete

a favorable outcome. To the best of our knowledge, this is the first report of a patient presenting with bilateral ocular symptoms due to SWDAVF with progressive bilateral ocular symptoms related to cranial nerve compression is warranted surgical intervention.

#### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

#### Financial support and sponsorship

Nil.

#### Conflicts of interest

There are no conflicts of interest.

#### REFERENCES

- Awad IA, Little JR, Akarawi WP, Ahl J. Intracranial dural arteriovenous malformations: Factors predisposing to an aggressive neurological course. *J Neurosurg* 1990;72:839-50.
- Ezura M, Takahashi A, Mizoi K. Dural arteriovenous shunts involving the sphenoparietal sinus: A case report. *Interv Neuroradiol* 1996;2:223-8.
- Fukuda H, Miyake K, Kunieda T, Murao K. Endovascular treatment of sphenoid wing dural arteriovenous fistula with pure cortical venous drainage. *J Stroke Cerebrovasc Dis* 2014;23:1730-5.
- Kandyba DV, Babichev KN, Stanishevskiy AV. Dural arteriovenous fistula in the sphenoid bone lesser wing region: Endovascular adjuvant techniques of treatment and literature review. *Interv Neuroradiol* 2018;24:559-66.
- Khadavi NM, Mancini R, Nakra T. Rare dural arteriovenous fistula of the lesser sphenoid wing sinus. *Ophthalmic Plast Reconstr Surg* 2009;25:404-6.
- Kim MJ, Shin YS, Ihn YK. Transvenous embolization of cavernous and paracavernous dural arteriovenous fistula through the facial vein: Report of 12 cases. *Neurointervention* 2013;8:15-22.
- Misaki K, Uchiyama N, Mohri M. Sphenoid wing dural arteriovenous fistula with ocular symptoms. *World Neurosurg* 2017;97:753-3.
- Murakami T, Nakamura H, Nishida T. Dural arteriovenous fistula in a sinus of the lesser sphenoid wing: A case report. *NMC Case Rep J* 2017;4:47-50.
- Nakajima H, Ishiguro T, Terada A, Komiyama M. Dural arteriovenous fistula of the sinus of the lesser sphenoid wing presenting with Pontine hemorrhage. *World Neurosurg* 2017;98:17-21.
- Nomura S, Anegawa S, Nakagawa S. Subarachnoid hemorrhage caused by dural arteriovenous fistula of the sphenobasal sinus-case report. *Neurol Med Chir (Tokyo)* 2002;42:255-8.

11. Osburn JW, Kim LJ, Spetzler RF. Aberrant venous drainage pattern in a medial sphenoid wing dural arteriovenous fistula: A case report and review of the literature. *World Neurosurg* 2013;80:e381-6.
12. Park JW, Lee JY. Traumatic intracerebral and subarachnoid hemorrhage due to a ruptured pseudoaneurysm of middle meningeal artery accompanied by a medial sphenoid wing dural arteriovenous fistula. *Korean J Neurotrauma* 2017;13:162-6.
13. Rezende MT, Piotin M, Mounayer C, Spelle L, Abud DG, Moret J. Dural arteriovenous fistula of the lesser sphenoid wing region treated with onyx: Technical note. *Neuroradiology* 2006;48:130-4.
14. Ruiz DS, Fasel JH, Rufenacht DA, Gailloud P. The sphenoparietal sinus of breschet: Does it exist? An anatomic study. *AJNR Am J Neuroradiol* 2004;25:112-20.
15. Sarma D, ter Brugge K. Management of intracranial dural arteriovenous shunts in adults. *Eur J Radiol* 2003;46:206-20.
16. Shi ZS, Ziegler J, Feng L, Gonzalez NR, Tateshima S, Jahan R, *et al.* Middle cranial fossa sphenoidal region dural arteriovenous fistulas: Anatomic and treatment considerations. *AJNR Am J Neuroradiol* 2013;34:373-80.
17. Tanaka T, Kato N, Arai T, Hasegawa Y, Abe T. Surgical treatment of a sylvian-middle fossa dural arteriovenous fistula draining into the basal vein of Rosenthal with frontotemporal craniotomy. *J Stroke Cerebrovasc Dis* 2012;21:333-7.
18. Tanoue S, Kiyosue H, Okahara M, Sagara Y, Hori Y, Kashiwagi J. Para-cavernous sinus venous structures: Anatomic variations and pathologic conditions evaluated on fat-suppressed 3D fast gradient-echo MR images. *AJNR Am J Neuroradiol* 2006;27:1083-9.
19. Thompson BG, Doppman JL, Oldfield EH. Treatment of cranial dural arteriovenous fistulae by interruption of leptomeningeal venous drainage. *J Neurosurg* 1994;80:617-23.
20. Tubbs RS, Salter EG, Wellons JC 3<sup>rd</sup>, Blount JB, Oakes WJ. The sphenoparietal sinus. *Neurosurgery* 2007;60 Suppl 1:ONS9-12; discussion ONS12.
21. van Rooij WJ, Sluzewski M. Curative embolization with onyx of dural arteriovenous fistulas with cortical venous drainage. *AJNR Am J Neuroradiol* 2010;31:1516-20.
22. Watanabe J, Maruya J, Nishimaki K. Surgical treatment of a dural arteriovenous fistula in the sphenoid wing with a unique drainage pattern through the basal vein of Rosenthal. *NMC Case Rep J* 2015;2:88-92.
23. Zhou LF, Chen L, Song DL, Gu YX, Leng B. Dural arteriovenous fistula of the sphenobasilar sinus with concomitant meningioma: Case report and review of the literature. *Neurosurg Rev* 2007;30:269-74.

**How to cite this article:** Shimizu Y, Tokuda K, Park C. Sphenoid wing dural arteriovenous fistula: A case report and literature review. *Surg Neurol Int* 2020;11:438.