

Isolated Oculomotor Nerve Palsy Caused by Cavernous Sinus Dural Arteriovenous Fistula: Case Report¹

해면정맥동 경막동정맥루에 의한 고립성 동안신경마비: 증례 보고¹

Yon Kwon Ihn, MD¹, Won Sang Jung, MD¹, Bum-Soo Kim, MD²

¹Department of Radiology, St. Vincent's Hospital, College of Medicine, The Catholic University of Korea, Suwon, Korea

²Department of Radiology, Seoul St. Mary's Hospital, College of Medicine, The Catholic University of Korea, Seoul, Korea

Cavernous dural arteriovenous fistula (DAVF), which usually presents with conjunctival injection, proptosis, loss of visual acuity, and ophthalmoplegia, is a rare cause of ophthalmoplegia. Thus, it may be overlooked when the typical symptoms are lacking. There have been some cavernous DAVF case reports presenting with isolated oculomotor, abducens and trochlear nerve palsy. We report a patient presenting with isolated oculomotor palsy, caused by cavernous DAVF, which was treated by transvenous coil embolization. This case suggests that cavernous DAVF should be considered in the differential diagnosis of isolated oculomotor nerve palsy and for which case - selective angiography and embolization may be helpful in reaching a diagnosis and providing a guide for optimal treatment.

Index terms

Cavernous Sinus
Oculomotor Palsy
Dural Arteriovenous Fistula

INTRODUCTION

Isolated oculomotor palsy is a rare symptom seen in patients with many, different neurological disorders (1, 2). It is often the first or even the sole manifestation of a pathology that potentially affects the nervous system. When older patients are presented with isolated ocular motor nerve palsy, without congestive ocular features, this condition is easily misdiagnosed as idiopathic, ischemic mononeuropathy or aneurysm. While isolated ocular motor nerve palsy, presenting as the only manifestation of cavernous dural arteriovenous fistula (DAVF), has occasionally been reported (3-5), we report a case of cavernous DAVF presenting with unusual, isolated oculomotor nerve palsy and treated by transvenous coil embolization.

CASE REPORT

A 76-year-old woman was presented with the sudden onset

of left-sided ptosis and diplopia. The patient had hypertension during the past twenty years. She did not complain of any ocular pain or conjunctival injection. On examination, she had left-sided ptosis, a mildly dilated pupil with a sluggish light reflex, and impaired medial, upward gaze of her left eye. Her visual acuity and ocular fundus appeared normal. To rule out the possibility of subarachnoid hemorrhage or an aneurysm, CT angiography was performed. It revealed no aneurysm, although an axial CT source image showed asymmetric contrast enhancement in the left cavernous sinus (Fig. 1A). Following a diffusion-weighted MRI, it showed no mid-brain infarction. However, contrast enhanced, dynamic magnetic resonance angiography (MRA) revealed rapid arterial phase filling of the left cavernous and ipsilateral inferior petrosal sinuses (Fig. 1B). Digital subtraction angiography (DSA) of the left internal carotid artery demonstrated a parasellar vascular blush. Shunted contrast pooled in the supero-posterior cavernous sinus, and

Received May 22, 2012; Accepted July 8, 2012

Corresponding author: Yon Kwon Ihn, MD

Department of Radiology, St. Vincent's Hospital,
College of Medicine, The Catholic University of Korea,
93 Jungbu-daero, Paldal-gu, Suwon 442-723, Korea.
Tel. 82-31-249-7491 Fax. 82-31-247-5713
E-mail: ihn@catholic.ac.kr

Copyrights © 2012 The Korean Society of Radiology

drained into the ipsilateral inferior petrosal sinus (Fig. 1C). DSA of the left external carotid artery showed a shunt from the recurrent meningeal artery to the cavernous sinus (Fig. 1D). No anterior drainage was observed through the superior ophthalmic vein. Endovascular treatment, under local anesthesia, was then performed five days after the diagnostic angiography. The patient was administered a bolus of intravenous heparin (50 IU/kg body weight) to achieve an activated clotting time of greater than 250 seconds. We accessed the left common femoral artery, after which, a 5-Fr catheter (Bern; Boston Scientific, Natick, MA, USA) was placed in the left external carotid artery. A 6-F guide catheter (Envoy, Cordis Endovascular, Miami Lakes, FL, USA) was then placed in the left internal jugular vein, via a

femoral venous sheath. A Prowler 18 microcatheter (Cordis endovascular, Miami Lakes, FL, USA) was prepared and passed into the left inferior petrosal sinus over an Agility 14 soft microguidewire (Cordis Endovascular, Miami Lakes, FL, USA). The microcatheter was navigated to the superoposterior aspect of the left cavernous sinus and a total of two detachable coils (GDC 10; Boston Scientific, Natick, MA, USA) and undetectables coils (Tornado coils; Cook, Bloomington, IN, USA) were used to perform coil embolization. A post-embolization angiogram demonstrated near complete occlusion of the cavernous DAVF (Fig. 1E). Following embolization, patient's ptosis and diploic symptom were improved for one week, although, she still complained of subtle diplopia, which, however, was subse-

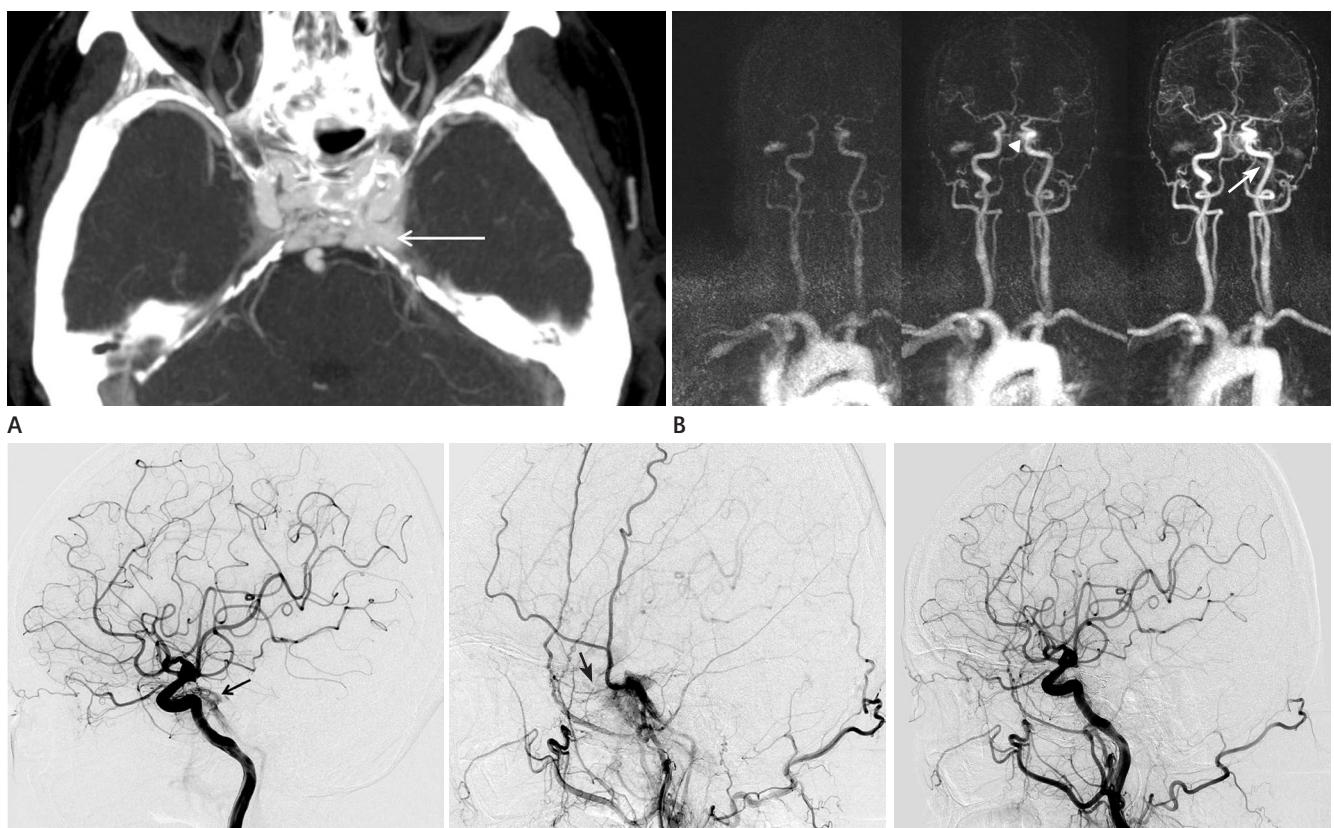


Fig. 1. A 76-year-old female patient with a cavernous dural arteriovenous fistula.

A. An axial CT source image shows asymmetrical contrast filling in the left cavernous sinus, especially, in the posterior aspect of the cavernous sinus (arrow).

B. Time-resolved contrast MRA images show a rapid arterial phase filling of both the left cavernous sinus (arrowhead) and the inferior petrosal sinus (arrow) because of the left cavernous dural arteriovenous fistula.

C. Left internal carotid angiography shows early filling of the superoposterior cavernous sinus (arrow) with drainage into the inferior petrosal sinus. Note that the superior ophthalmic vein was not visualized.

D. Left external carotid angiography shows a cavernous dural arteriovenous fistula, supplied by a recurrent meningeal artery (arrow) from the middle meningeal artery, and with drainage into the inferior petrosal sinus.

E. Left common carotid angiography demonstrates complete occlusion of the fistula.

Note.—MRA = magnetic resonance angiography

quently improved. Two months after embolization, her ptosis had disappeared and there was no diploic symptom.

DISCUSSION

Cavernous sinus DAVF is a communication between the cavernous sinus and the dural branches of the internal carotid artery, external carotid artery or both. Compared with the transverse-sigmoid sinus, which is the most frequent site of occurrence in Western patients, the cavernous sinus is the most common location of DAVFs in Asians (6). Cavernous DAVF is known to have a favorable prognosis and a spontaneous regression rate of 10-73%. When the shunts drain posteriorly into the inferior petrosal sinus, isolated oculomotor, trochlear, or abducens nerve palsy may develop (3).

The cavernous DAVF in our patient was not suspected clinically as the patient lacked the congestive orbito-ocular features of more typical, anterior draining fistulas ("red-eyed shunt") (3). MRA and DSA imaging correlates with red-eyed shunts, i.e. dilated superior or inferior ophthalmic veins and extraocular muscles, which were absent in our patient since most of the blood drained posteriorly.

It is still unclear why the oculomotor nerve is the most commonly compromised nerve in cavernous DAVF with posterior drainage. Various mechanisms have been proposed as the cause of isolated ocular motor nerve palsy, including nerve compression, vascular steal, and venous congestion (5). The most plausible cause of oculomotor nerve palsy is compression of a nerve by the distended superoposterior aspect of the cavernous sinus against the roof of cavernous sinus. Miyachi et al. (7) reported that the oculomotor nerve pierces the cavernous sinus roof, lateral to the posterior clinoid process, and courses through the upper part of the lateral wall. Therefore, superoposterior drainage of venous flow can compress the oculomotor nerve against the roof of the cavernous sinus. However, as the entrance point of the oculomotor nerve into the cavernous sinus and neurovascular relationships of other cranial nerves in the cavernous sinus are so variable, and oculomotor nerve palsy may occur prior to abducens nerve palsy in some cases. The alternative mechanism of oculomotor nerve palsy is dural arterial steal, which is caused by blood shunting from the meningohypophyseal or inferolateral trunk that supplies the intracavernous part of the oculo-

motor nerve, to the cavernous sinus (5, 8).

Although the diagnosis of DAVF may commonly be suspected on planar CT and MR imaging, DSA is still necessary in order to confirm the diagnosis and to define the lesion. A recent study suggested that dynamic contrast MRA can diagnose and classify a cranial DAVF, using a commercially available form of time-resolved MRA (trMRA) at 3T. However, the use of trMRA in the area of cranial DAVF is recommended for the purpose of screening and surveillance, and not as a tool for the complete characterization of a fistula (9).

The treatment options for cavernous DAVF include manual compression of the ipsilateral carotid artery, transarterial embolization of feeder vessels, and transvenous embolization, the last of which is generally considered to be an effective method treatment for cavernous DAVF. The transfemoral venous approach reaches the cavernous sinus through the inferior petrosal sinus, and is the preferred method, due to its anatomically short and straight (10).

The outcome of oculomotor nerve palsy with cavernous DAVF has generally been favorable, regardless of the treatment method. This may have been due to the low shunt pressure, which is more responsive to the treatment than high-pressure shunts and often resolves spontaneously (7). Furthermore, treatment using embolization leads to more rapid resolution of clinical manifestation (3). Klisch et al. (10) reported that in their study, overpacking of the cavernous sinus or thrombosis of the cavernous sinus may have been a cause of the paradoxical worsening of their cranial nerve symptoms. However, the management of these complications by conservative analgesics and steroid therapy can result in successful clinical outcomes.

In conclusion, review of the causes of isolated oculomotor nerve palsy suggests that there can be numerous causes. We propose that carotid DAVF should be considered in the differential diagnosis of isolated oculomotor nerve palsy, and in which case, contrast-enhanced MRA and cerebral angiography may be helpful for both making the correct diagnosis and providing the appropriate treatment.

REFERENCES

- Chen PR, Amin-Hanjani S, Albuquerque FC, McDougall C, Zabramski JM, Spetzler RF. Outcome of oculomotor nerve

- palsy from posterior communicating artery aneurysms: comparison of clipping and coiling. *Neurosurgery* 2006; 58:1040-1046; discussion 1040-1046
2. Kobayashi H, Kawabori M, Terasaka S, Murata J, Houkin K. A possible mechanism of isolated oculomotor nerve palsy by apoplexy of pituitary adenoma without cavernous sinus invasion: a report of two cases. *Acta Neurochir (Wien)* 2011;153:2453-2456; discussion 2456
3. Acierno MD, Trobe JD, Cornblath WT, Gebarski SS. Painful oculomotor palsy caused by posterior-draining dural carotid cavernous fistulas. *Arch Ophthalmol* 1995;113:1045-1049
4. Kurata A, Takano M, Tokiwa K, Miyasaka Y, Yada K, Kan S. Spontaneous carotid cavernous fistula presenting only with cranial nerve palsies. *AJNR Am J Neuroradiol* 1993; 14:1097-1101
5. Wu HC, Ro LS, Chen CJ, Chen ST, Lee TH, Chen YC, et al. Isolated ocular motor nerve palsy in dural carotid-cavernous sinus fistula. *Eur J Neurol* 2006;13:1221-1225
6. Kim MS, Han DH, Kwon OK, Oh CW, Han MH. Clinical characteristics of dural arteriovenous fistula. *J Clin Neurosci* 2002;9:147-155
7. Miyachi S, Negoro M, Handa T, Sugita K. Dural carotid cavernous sinus fistula presenting as isolated oculomotor nerve palsy. *Surg Neurol* 1993;39:105-109
8. Hawke SH, Mullie MA, Hoyt WF, Hallinan JM, Halmagyi GM. Painful oculomotor nerve palsy due to dural-cavernous sinus shunt. *Arch Neurol* 1989;46:1252-1255
9. Farb RI, Agid R, Willinsky RA, Johnstone DM, Terbrugge KG. Cranial dural arteriovenous fistula: diagnosis and classification with time-resolved MR angiography at 3T. *AJNR Am J Neuroradiol* 2009;30:1546-1551
10. Klisch J, Huppertz HJ, Spetzger U, Hetzel A, Seeger W, Schumacher M. Transvenous treatment of carotid cavernous and dural arteriovenous fistulae: results for 31 patients and review of the literature. *Neurosurgery* 2003;53:836-856; discussion 856-857

해면정맥동 경막동정맥루에 의한 고립성 동안신경마비: 증례 보고¹

인연권¹ · 정원상¹ · 김범수²

해면정맥동에 발생하는 경막동정맥루는 결막충혈, 안구돌출, 시력장애와 안구마비 등을 일으킬 수 있다. 하지만, 전형적인 안구증상 없는 고립성 동안신경마비로 발현되는 해면정맥동 경막동정맥루는 드물다. 저자들은 76세 여자 환자에서 발생한 고립성 동안신경마비의 원인인 해면정맥동 경막동정맥루의 경정맥치료의 경험을 보고하고자 한다.

¹가톨릭대학교 의과대학 성빈센트병원 영상의학과, ²가톨릭대학교 의과대학 서울성모병원 영상의학과