

Isolated Bilateral Abducens Nerve Palsy due to Carotid Cavernous Dural Arteriovenous Fistula

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Carotid cavernous dural arteriovenous fistula (DAVF) usually presents with conjunctival injection, proptosis, loss of visual acuity and ophthalmoplegia. There have been some carotid cavernous DAVF case reports presenting with isolated oculomotor, abducens and trochlear nerve palsy. We experienced a patient presenting with bilateral abducens nerve palsy and no other ocular signs who was diagnosed as carotid cavernous DAVF after conventional angiography. According to this case, carotid cavernous DAVF should be considered in the differential diagnosis of isolated bilateral abducens nerve palsy, in which case conventional angiography may be helpful in diagnosis.

Carotid cavernous DAVFs are abnormal communications between the carotid arterial system and the cavernous sinus. The common symptoms of carotid cavernous DAVFs are conjunctival injection, proptosis, loss of visual acuity, ophthalmoplegia and orbital bruit. If there are typical ocular signs, it is not difficult to suspect a carotid cavernous DAVF as the cause. There have been carotid cavernous DAVF cases with ophthalmoplegia without any other typical ocular signs, and in these cases it has not always been easy to diagnose the carotid cavernous DAVF (Sempere *et al.* 1991; Selky and Purvin, 1994; Kwak *et al.* 1995). We experienced an unusual carotid cavernous DAVF case presenting with bilateral abducens nerve palsy and no other ocular signs.

CASE REPORT

A 58-year-old woman developed sudden horizon-

Received February 25, 1998

Accepted April 17, 1998

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tal diplopia. One month before she was admitted to our hospital, she had been diagnosed with hypertension and a mild degree of mitral regurgitation at a private clinic. There was no past history of diabetes. She did not complain of any ocular pain. Clinical evaluation showed impaired abductive movement bilaterally, especially in the left eye. There was no proptosis, conjunctival injection or orbital bruit. Both pupils were isocoric and immediately reactive to light. Her visual acuity and ocular fundus appeared normal. The remainder of the neurological examination was normal. A gadolinium-enhanced brain MRI showed no abnormalities. Digital subtraction angiography (DSA) demonstrated a left side carotid cavernous DAVF supplied by the left internal carotid artery (ICA) meningeal branch, with drainage into the right cavernous sinus via the intercavernous sinuses and right inferior petrosal sinus (Fig. 1). There was no external carotid artery contribution to the fistula.

Left cavernous sinus embolization with a platinum coil was performed by transvenous approach. One week after embolization there was improvement in the abductive movement of both eyes, especially the right eye. One month after embolization the

patient was readmitted to our department for follow-up angiography. She complained of tinnitus in her right ear. There was no limitation of abductive movement in both her eyes. Her diplopic symptom was improved after embolization but she complained of subtle diplopia, especially in distance vision,

which was later improved. Follow-up DSA demonstrated the remaining flow into the right cavernous sinus and the right inferior petrosal sinus. No further intervention was performed and she was just recommended for intermittent carotid artery compression. Six months after embolization, tinnitus of her right

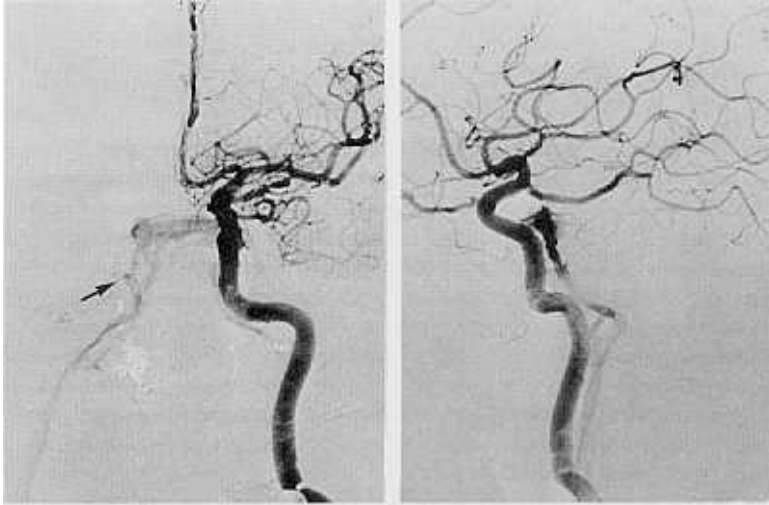


Fig. 1. Left carotid cavernous dural arteriovenous fistula draining into the right cavernous sinus via intercavernous sinuses and right inferior petrosal sinus (arrow) in AP and lateral digital subtraction angiography of the left carotid artery.

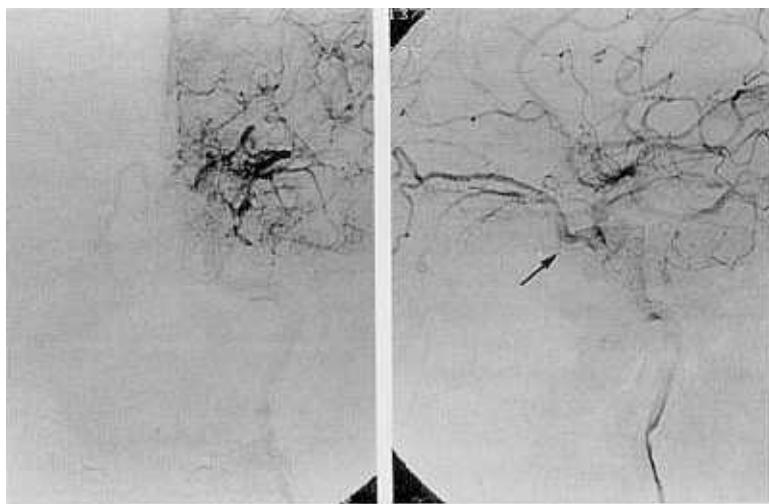


Fig. 2. Left carotid cavernous dural arteriovenous fistula mainly draining into the left superior ophthalmic vein (arrow). There was reduced flow to the right inferior petrosal sinus compared to the previous angiography.

ear had disappeared and there was no diplopic symptom. On physical examination there was no limitation of movement in both eyes and no ptosis, but mild conjunctival injection was found especially on her left eye. Follow-up DSA demonstrated that there was decreased flow into the right inferior petrosal sinus and a newly-developed tortuous and dilated left superior ophthalmic vein compared to previous angiography (Fig. 2). Because there were no symptoms, the patient was discharged without further intervention.

DISCUSSION

Carotid cavernous DAVFs are classified by several methods. Pathogenetically they are classified into spontaneous and traumatic fistulas, hemodynamically into high flow and low flow fistulas and angiographically into four types (Barrow *et al.* 1985). Tears of the ICA, mainly due to trauma, cause direct fistulas. They usually lie in the anterior part of the cavernous sinus and drain into the orbit via the ophthalmic venous system. Therefore they usually present with chemosis, proptosis, loss of visual acuity and ophthalmoplegia. Spontaneous fistulas usually present less severe symptoms and signs than traumatic fistulas, since most fistulas contain blood flowing at a low rate. Sometimes spontaneous dural fistulas lie in the posterior part of the cavernous sinus and drain posteriorly into the inferior petrosal sinus. In that case they present with mild ocular signs because of a low flow rate and the direction of the venous outflow (Leonard *et al.* 1984; Gregory *et al.* 1988; Kurata *et al.* 1993; Acierno *et al.* 1995).

In this case, the presenting symptoms and signs were unusual. There were no typical ocular signs like proptosis, conjunctival injection or bruit. After initial angiography we found that there was no flow to the superior ophthalmic vein, so we thought there was the possibility of no communication between the cavernous sinus and the superior ophthalmic vein (Dandy and Follis, 1941). But follow-up DSA showed communication between the cavernous sinus and the superior ophthalmic vein. There have been some cases like ours presenting with the direction of venous outflow changing from the inferior pet-

rosal sinus to the superior ophthalmic vein (Hawke *et al.* 1989). So we speculated that the paucity of ocular signs in our patient was due to the location of dural fistulas. These fistulas lie in the posterior of the cavernous sinus and the main cavernous venous outflow is drained posteriorly by the petrosal sinus according to the pressure gradient, so there is no pressure effect to the orbital venous outflow.

Bilateral abduction failure was another unusual feature of our case. In carotid cavernous DAVFs, ocular signs are usually in the side of the lesion, but there have been cases presenting with both or contralateral ocular signs (Graham, 1966; Bynke and Efsing, 1970; Gregory *et al.* 1988; Jun *et al.* 1997). It can be explained by a venous drainage system of the cavernous sinus. There is communication between both sides of the cavernous sinus so pressure can be transmitted to the contralateral sinus, and if the cavernous sinus and the superior ophthalmic vein are freely communicating, then typical ocular signs can appear. The proposed mechanisms of ophthalmoplegia in carotid cavernous DAVFs are swelling of the extraocular muscles due to venous congestion, ischemia of the cranial nerve by vascular steal, and compression of the cranial nerve by distended sinus (Leonard *et al.* 1984).

In our case, there was no evidence of swollen extraocular muscles by MR imaging. The possibility of nerve ischemia by vascular steal is low because there was no other cranial nerve involvement and no contribution to the fistula from the right ICA which supplies the right side cranial nerves. So we speculated that the cause of contralateral right eye abduction failure might be compression of the abducens nerve by distended right inferior petrosal sinus against the petroclinoid ligament (Leonard *et al.* 1984).

In a review of the cases of bilateral abducens nerve palsy, the causes are numerous, including tumor, trauma, infection, demyelinating disease and subarachnoid hemorrhage etc (Kearne, 1976). We propose that carotid cavernous DAVFs should be considered in the differential diagnosis of isolated bilateral abducens nerve palsy, in which case cerebral angiography may be helpful in making the correct diagnosis, especially if there are no gross abnormalities in MRI, and in providing the proper treatment.

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