

Case Report

Isolated Oculomotor Nerve Palsy in a White-Eyed Patient with Dural Carotid-Cavernous Sinus Fistulas: A Case Report

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The study reported a woman with dural carotid-cavernous sinus fistulas (CCFs) who presented with a unilateral white-eyed appearance, and painful oculomotor nerve palsy with pupillary involvement. After cerebral angiography, which revealed posterior drainage of the fistulas, the dural CCFs closed, the oculomotor nerve palsy subsided spontaneously and no recurrence occurred throughout the 2-year follow-up. Dural CCFs should be kept in mind when patients present with painful oculomotor nerve palsy. Moreover, cerebral angiography remains the standard diagnostic method and fistulas may close spontaneously following this procedure.

Keywords: Dural carotid-cavernous sinus fistulas, Oculomotor nerve palsy, Diplopia

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The most common and most serious cause of isolated painful oculomotor palsy is posterior communicating artery aneurysm. However, differential diagnoses to be considered are neoplasm, inflammation, carotid-cavernous sinus thrombosis and carotid cavernous sinus fistulas (CCFs)⁽¹⁻⁴⁾.

The present study reports a patient with headache and diplopia from painful oculomotor nerve palsy caused by posterior-draining dural CCFs.

Case Report

A sixty-three-year-old woman with underlying diabetes mellitus and hypertension complained of intermittent left-sided orbitofrontal headache for 4 months. Two weeks prior, she was investigated by computed tomography of the brain at another hospital, and was diagnosed with ischemic stroke. She was prescribed antiplatelet medication. One week later, she developed left periocular pain and binocular diplopia associated with blurred vision of the left eye. On ocular examination, the best-corrected visual acuity and fundi of both eyes were normal. There was mild ptosis, semi dilatation, and sluggish reactivity to light of her left

pupil. The extra-ocular eye movement examination disclosed impairment in adduction and infraduction of the left eye (Fig. 1). The red glass test detected diplopia with exotropia and left hypertropia of 3 and 8 prism diopters, respectively. Carotid bruit and conjunctival injection were not detected. The first impression was left partial oculomotor nerve palsy with pupillary involvement.

Therefore, magnetic resonance imaging (MRI) and magnetic resonance angiography (MRA) were done to detect the posterior communicating artery aneurysm. The abnormal MRI finding was abnormal flow void of the left-sided cavernous sinus in the T2-



Fig. 1 Left third cranial nerve palsy. Patient had mild ptosis and exotropia with limitation of adduction and infraduction of the left eye

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weighted image (Fig. 2). Dural CCFs were detected by MRA (Fig. 3). Cerebral angiography was performed, and disclosed dural arteriovenous fistulas at the posterior part of the left cavernous sinus, supplied by the left more than the right meningeal branches of the cavernous part of the internal carotid artery, the branches of the external carotid artery and venous drainage into the left inferior petrosal sinus (Fig. 4).

Spontaneous improvement of headache and eye movement after the first angiography (Fig. 5), along with subsequent angiography, confirmed closure of the fistulas (Fig. 6). Observation and an oral antiplatelet were ordered for the patient. During the 2-year follow-

up, no recurrence of the fistulas or painful oculomotor nerve palsy was found.

Discussion

In the present report, the patient had symptoms and signs of painful, incomplete oculomotor nerve palsy with pupillary involvement from posterior draining dural CCFs. Like the other studies^(1,2,4), headache or ipsilateral orbital pain and diplopia were the principal manifestations reported in patients with white-eyed CCFs. Moreover, almost 50% of dural CCFs

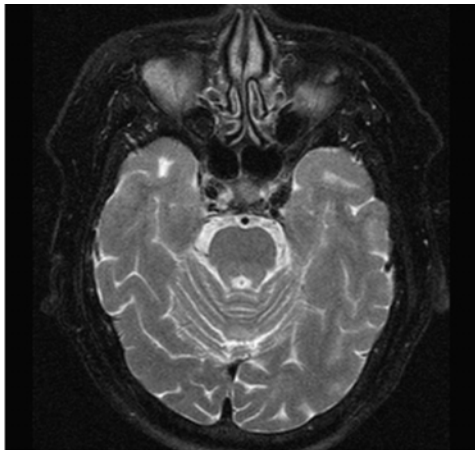


Fig. 2 MRI shows abnormal flow void of the left-sided cavernous sinus in T2-weighted image

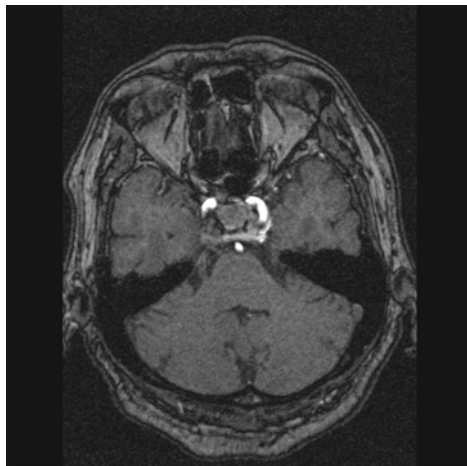


Fig. 3 MRA shows abnormal flow-related enhancement in the left-sided cavernous sinus crossing to the right side

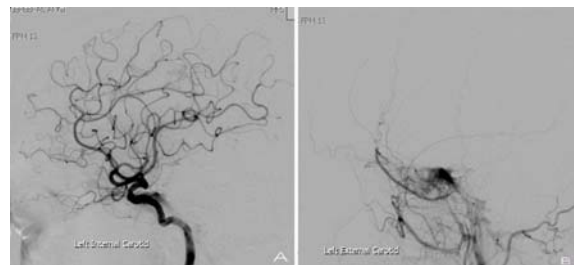


Fig. 4 Cerebral angiography shows (A) posterior draining dural CCFs supplied by branches of internal carotid artery, and (B) branches of external carotid artery and draining into the inferior petrosal sinus



Fig. 5 Normal extraocular eye movement was obtained after cerebral angiography

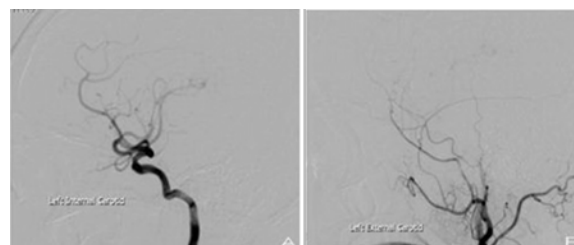


Fig. 6 Subsequent angiography confirmed complete resolution of the dural CCFs

closed spontaneously after angiography or after air flight travel, and the symptoms and signs began to resolve within days to weeks after angiography^(1,5-7). When the drainage route of the fistula was posterior, there were usually no symptoms or evidence of orbital congestion and in patients presenting with ocular motor nerve palsy, the oculomotor nerve was most often affected^(5,6,8). The signs of oculomotor nerve palsy could be complete with pupillary involvement, incomplete with pupillary involvement, or incomplete with pupillary sparing^(5,6). Various mechanisms have been proposed for cranial neuropathy, including neural compression, venous congestion or thrombosis and vascular steal, but the exact cause is unknown⁽¹⁻³⁾. MRA could be beneficial in the diagnosis of fistulas, although the gold standard diagnostic test is still cerebral angiography^(5,6,9,10) which not only additionally differentiates the CCFs from the life-threatening posterior communicating internal carotid artery aneurysm, but fistulas could also spontaneously close after the angiographic procedure.

In summary, though uncommon, dural CCFs should be considered in patients with painful oculomotor nerve palsy. MRI and MRA are useful investigative methods in detecting fistulas. However, cerebral angiography is still the gold standard diagnostic method. Moreover, almost 50% of dural CCFs close spontaneously after angiography or after air flight travel, and the symptoms and signs begin to resolve within days to weeks after angiography.

Potential conflicts of interest

None.

References

1. Hawke SH, Mullie MA, Hoyt WF, Hallinan JM, Halmagyi GM. Painful oculomotor nerve palsy due to dural-cavernous sinus shunt. *Arch Neurol* 1989; 46: 1252-5.
2. Miyachi S, Negoro M, Handa T, Sugita K. Dural carotid cavernous sinus fistula presenting as isolated oculomotor nerve palsy. *Surg Neurol* 1993; 39: 105-9.
3. 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: 1097-101.
4. Acierno MD, Trobe JD, Cornblath WT, Gebarski SS. Painful oculomotor palsy caused by posterior-draining dural carotid cavernous fistulas. *Arch Ophthalmol* 1995; 113: 1045-9.
5. Miller NR. Diagnosis and management of dural carotid-cavernous sinus fistulas. *Neurosurg Focus* 2007; 23: E13.
6. Miller NR. Carotid-cavernous sinus fistulas. In: Miller NR, Newman NJ, Biouesse V, Kerrison JB, editors. *Walsh and Hoyt's clinical neuro-ophthalmology*. Philadelphia: Lippincott Williams & Wilkins; 2005: 2283-92.
7. Grove AS Jr. The dural shunt syndrome. Pathophysiology and clinical course. *Ophthalmology* 1984; 91: 31-44.
8. Eggenberger E, Lee AG, Forget TR Jr, Rosenwasser R. A brutal headache and double vision. *Surv Ophthalmol* 2000; 45: 147-53.
9. Lv X, Jiang C, Li Y, Yang X, Wu Z. Isolated oculomotor nerve palsy in interventional neuroradiology. *Eur J Radiol* 2010; 74: 441-4.
10. Uehara T, Tabuchi M, Kawaguchi T, Mori E. Spontaneous dural carotid cavernous sinus fistula presenting isolated ophthalmoplegia: evaluation with MR angiography. *Neurology* 1998; 50: 814-6.

รายงานผู้ป่วยเส้นประสาทสมองที่สามเป็นอัมพาตเนื่องจากภาวะเชื่อมต่อของหลอดเลือดแดงกับหลอดเลือดดำ (dural carotid-cavernous sinus fistulas) โดยไม่มีอาการตาแดงนำมาก่อน

พัชรพิมพ์ มัทยาอนนท์

รายงานผู้ป่วยหญิงไทยที่ได้รับการวินิจฉัยเป็น dural carotid cavernous sinus fistulas ซึ่งมาด้วยอาการปวดศีรษะร่วมกับมีเส้นประสาทสมองที่ 3 เป็นอัมพาตโดยผู้ป่วยไม่มีตาแดงร่วมด้วย การฉีดสีวินิจฉัยพบว่า fistulas ดังกล่าวมีการระบายเลือดไปข้างหลัง และภาวะ fistulas ร่วมกับเส้นประสาทสมองที่ 3 เป็นอัมพาตหายเองภายหลังจากผู้ป่วยได้รับการฉีดสีวินิจฉัย นอกจากนี้ยังไม่พบการเป็นซ้ำตลอดระยะเวลาติดตามผู้ป่วยนาน 2 ปี ภาวะ dural carotid cavernous sinus fistulas เป็นภาวะที่ควรตระหนักถึงในผู้ป่วยที่มีอาการปวดศีรษะร่วมกับมีเส้นประสาทสมองที่ 3 เป็นอัมพาต การฉีดสีวินิจฉัยนอกจากจะเป็นการตรวจวินิจฉัย ที่มาตรฐานแล้วยังพบว่า fistulas สามารถปิดเองได้ภายหลังการฉีดสีวินิจฉัยด้วย
