CASE REPORTS

Sixth Nerve Palsy Secondary to Traumatic Carotid-Jugular Fistula

Kevin M. Halenda ^a, Scott Y. Rahimi^b, Jordan J. Patton^a, and Dilip A. Thomas^a

^aDepartment of Ophthalmology, Medical College of Georgia at Augusta University, Augusta, Georgia, USA; ^bDepartment of Neurosurgery, Medical College of Georgia at Augusta University, Augusta, Georgia, USA

ABSTRACT

Carotid-jugular fistulae are rare complications of penetrating head and neck trauma. We report an unusual case of an external carotid-internal jugular fistula presenting with diplopia due to sixth nerve paresis. A 38-year-old Caucasian female presented in the setting of acute neurovascular neck trauma and weeks later developed symptomatic diplopia. An acquired carotid-internal jugular fistula affecting the abducens nerve secondarily via compression from a distended inferior petrosal sinus was diagnosed and treated using endovascular coil embolisation, resolving the patient's symptoms. A posteriorly draining external carotid artery-internal jugular venous fistula can be an uncommon cause of a compressive sixth cranial nerve palsy.

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Case report

The sixth cranial nerve runs a long intracranial course originating in the dorsal pons and terminating on the ipsilateral lateral rectus muscle, rendering it susceptible to injury at numerous loci. Common mechanisms of sixth cranial nerve paresis include ischaemia, trauma, compression, and stretch injury. Herein, we report an unusual case of sixth nerve paresis caused by compression from a distended inferior petrosal sinus which resulted from the shunting of an external carotid-internal jugular arteriovenous fistula.

A 38-year-old female originally presented to our institution after sustaining a self-inflicted gunshot wound to the left anterior neck. Head and neck CT imaging identified fractures of the left mastoid tip with retained bullet fragment, inferior anterolateral occipital bone, left C2 transverse process, as well as a small subdural haematoma, multiple soft tissue haematomas, dissection of the left internal carotid artery, and loss of contrast opacification of the left external carotid artery, internal jugular vein, jugular bulb, sigmoid, and transverse sinuses. Subsequent imaging demonstrated an ischaemic infarct of the left lentiform nucleus and left sigmoid sinus thrombosis. After evaluation by endovascular neurosurgery, the patient was managed

conservatively with anticoagulation and ultimately discharged to a domestic abuse safe house.

Six weeks later, the patient presented for follow-up care in the neurosurgery clinic, where she complained of a new “vibration” in her left neck. Subsequent imaging demonstrated a stable left internal carotid artery dissection at the level of C1-C2, as well as the interval development of a small internal carotid pseudoaneurysm at the level of C2, and fistulation between the proximal left external carotid artery and the internal jugular vein. Transfemoral 9-vessel cerebral angiography confirmed a high flow fistula between the left external carotid artery and the left internal jugular vein with evidence of retrograde reflux into the transverse, sigmoid, inferior petrosal, and cavernous sinuses (Figure 1A–G). Elective therapeutic endovascular embolisation of the fistula was scheduled, however the patient was subsequently lost to follow-up prior to surgery.

Two months later, the patient again presented to the emergency department with new onset diplopia and left periorbital pain following facial trauma. The patient reported striking her left orbit on the edge of a piece of furniture while walking with crutches. She was discharged after a non-contrasted maxillofacial CT study showed no acute findings.

Figure 1. Angiography and endovascular treatment of external-carotid-internal jugular fistula.

- a. Lateral angiogram demonstrating a high flow proximal ECA-IJ fistula from gunshot wound to neck.
- b. AP view of the high flow fistula with retrograde flow into the intracranial venous sinuses.
- c. Lateral angiogram showing retrograde reflux into the IPS and cavernous sinus. Normal ECA anatomy not seen due to shunting into the high flow fistula.
- d. Point of proximal ECA-IJ fistula measuring 12.6×10.4 mm.
- e. Intraoperative angiogram showing coil embolisation of the ECA-IJ fistula.
- f. AP angiogram showing coil mass in the fistula with opacification of the ICA and IJ.
- g. Lateral angiogram showing occlusion of the ECA-IJ fistula. There is some opacification of the normal ECA branches including the superior thyroid artery (arrow), lingual artery (double arrow) and internal maxillary artery (large arrow).

At her ophthalmology follow-up appointment 2 days later, she noted persistent blurry vision and horizontal binocular diplopia. On examination, pinhole visual acuity was 20/20 OS. Ocular motility testing demonstrated mild painful limitation of abduction OS. Alternate cover testing demonstrated an incomitant esodeviation measuring 9 PD in right gaze, 18 PD in primary gaze, and 35PD in left gaze. Apart from mild injection OS, the remainder of the exam was normal. She was referred to and seen by the neuro-ophthalmology service 2 weeks later at which time she reported

worsening diplopia with painful abduction, and headache. The previously noted examination findings were confirmed and stable. Furthermore, diminished saccadic velocity with abduction OS was noted, suggesting left sixth nerve paresis.

Upon review of the previous CT scan, a focal soft tissue density within the left anterior ethmoid sinus was noted without a clear fracture. Although the delayed onset of diplopia and saccadic slowing highly supported a left CN VI palsy, the prominent pain with abduction immediately after the reported subsequent orbital injury were felt to

warrant exploration to rule out an occult medial wall fracture, despite minimal CT findings.

The following day, a transcaruncular medial orbitotomy was performed. Intraoperative forced ductions prior to exploration were performed and did not suggest restriction. Upon exploration, a minor depression of the inferomedial orbital wall fracture without significant soft tissue incarceration to cause restriction was identified, thus confirming that the diplopia was truly due to abducens nerve paresis. Carotid dissection was considered as a possible etiology of the abducens palsy, however, given the delayed onset of symptoms and lack of associated Horner syndrome this seemed unlikely. Therefore, a left abducens nerve palsy secondary to compression from the inferior petrosal sinus, distended due to significant retrograde reflux from the left external carotid-internal jugular fistula, was diagnosed as the true cause of her symptoms.

Approximately 3 weeks later, the patient underwent endovascular coil embolisation of the left external carotid-internal jugular fistula by neurovascular surgery with complete occlusion of the fistula and resolution of retrograde reflux into

the intracranial sinus system. Though the patient has failed to report for subsequent ophthalmology follow-up, she noted complete resolution of her pain and diplopia on a subsequent phone interview. The patient underwent a repeat angiogram with neurosurgery 1 year after the original coil embolisation which demonstrated normal opacification of the left external carotid vessel. There continues to be evidence of a small dissecting pseudoaneurysm in the proximal internal carotid artery with no haemodynamic significance (Figure 2). Follow up angiography will be performed in the future to confirm stability of the pseudoaneurysm.

Discussion

Acquired carotid-jugular fistulae are rare complications of penetrating trauma to the neck.¹ Due to the high-flow arteriovenous nature of these fistulae, patients may present with a pulsatile neck mass, continuous thrill, bruit, tinnitus, headache, and signs of high-output heart failure. Although the combination of history and physical findings are often suggestive, conventional catheter or CT/MR angiography and ultrasound maybe used confirm

Figure 2. Follow-up angiography of external-carotid-internal jugular fistula. One year follow-up lateral angiogram showing normal opacification of internal carotid artery, all branches of the external carotid artery and no evidence of fistula or venous reflux.

the diagnosis of carotid-jugular fistulae. Treatment may be achieved with either open surgical ligation or more modern endovascular techniques.²

Carotid cavernous sinus fistulae (CCF) are aberrant communications between the carotid artery and the cavernous sinus. CCF are commonly divided into two clinical categories: direct and indirect (dural). A popular classification system outlined by Barrow et al further subdivides all CCF into four different types (A-D).³ Direct CCF, characterised by high flow communication between the internal carotid artery and the cavernous sinus, are usually traumatic in nature. In contrast, indirect dural CCF represent communications between the meningeal branches of the internal and/or external carotid artery and the cavernous sinus.

Both categories of CCF are capable of causing cranial neuropathies by shunting high-pressure arterial blood into the cranial venous sinus system.⁴ Direct CCF are more likely to be symptomatic and present with the classical triad of clinical symptoms featuring chemosis, ocular bruit, and pulsatile exophthalmos due to higher flow than their dural counterparts. Anteriorly-draining dural CCF may also be associated with orbital congestion, however cases of dural CCF draining posteriorly into the inferior petrosal sinuses are frequently asymptomatic or occasionally develop painful ocular motor nerve paresis.⁵

We report a rare case in which a direct external carotid-internal jugular fistula stemming from trauma resulted in an ipsilateral sixth nerve palsy. As with dural arteriovenous fistulae of the inferior petrosal sinus, cranial neuropathies can result from compression or distension from elevated intracranial pressure caused by increased venous pressure from retrograde shunting of arterial blood.⁶ In our case, digital subtraction angiography demonstrated significant retrograde reflux of high-pressure arterial blood into the transverse and sigmoid sinuses, which secondarily distended the inferior petrosal and cavernous sinuses. The high pressure transmitted into the inferior petrosal sinus by retrograde flow through the internal jugular vein was primarily responsible for the sixth nerve paresis in this case, as the patient lacked

clinical evidence of orbital congestion or other cranial neuropathies suggestive of cavernous sinus involvement. The isolated involvement of the ipsilateral abducens nerve in this case is most consistent with compression within Dorello's canal, where the nerve travels together with the inferior petrosal sinus before entering the cavernous sinus.⁷ Traumatic carotid-jugular fistulae are inherently rare entities and to our knowledge, this is the first reported case of a sixth nerve palsy resulting from this particular arteriovenous fistula.

Declaration of interest

The above authors have no conflicts of interest to disclose.

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ORCID

Kevin M. Halenda <http://orcid.org/0000-0003-3966-9660>

References

1. Feliciano DV. Management of penetrating injuries to carotid artery. *World J Surg.* 2001;25:1028–1035.
2. Ezemba N, Ekpe EE, Ezike HA, Anyanwu CH. Traumatic common carotid–jugular fistula: report of 2 cases. *Tex Heart Inst J.* 2006;33:81–83.
3. Barrow DL, Spector RH, Braun IF, Landman JA, Tindall SC, Tindall GT. Classification and treatment of spontaneous carotid-cavernous sinus fistulas. *J Neurosurg.* 1985;62(2):248–256. doi:10.3171/jns.1985.62.2.0248.
4. Brosnahan D, McFadzean RM, Teasdale E. Neuro-ophthalmic features of carotid cavernous fistulas and their treatment by endoarterial balloon embolisation. *J Neurol Neurosurg Psychiatry.* 1992;55:553–556.
5. Miller NR. Dural carotid-cavernous fistulas: epidemiology, clinical presentation, and management. *Neurosurg Clin N Am.* 2012;23(1):179–192. doi:10.1016/j.nec.2011.09.008.
6. Barnwell SL, Halbach VV, Dowd CF, Higashida RT, Hieshima GB. Dural arteriovenous fistulas involving the inferior petrosal sinus: angiographic findings in six patients. *AJNR Am J Neuroradiol.* 1990;11:511–516.
7. Mittal SO, Siddiqui J, Katirji B. Abducens nerve palsy due to inferior petrosal sinus thrombosis. *J Clin Neurosci.* 2017;40:69–71. doi:10.1016/j.jocn.2017.02.018.