Abducens nerve palsy and ipsilateral Horner syndrome as the presenting finding of carotid cavernous fistula

Case Presentation

An 84-year-old female with history of hypertension and hyperlipidemia presented with nontraumatic left eyelid edema, erythema, and ptosis followed by acute painless binocular horizontal diplopia. Best corrected visual acuity was 20/20 OD and 20/30 OS. The OD pupil measured 4 mm in the dark and 2 mm in the light, and the OS pupil measured 3 mm in the dark and 2 mm in the light. No relative afferent pupillary defect was detected. There was a 50 prism diopter esotropia in primary gaze and an abduction deficit of −4 OS (Fig. 1). The OD moved normally. Intraocular pressure measured 17 mm Hg OD and 16 mm Hg OS. Anterior segment examination of OD was normal, while OS showed dilated and tortuous deep episcleral vessels with arterialization of vessels. Fundus examination showed no disc edema, and the macula and vessels looked normal. There was no proptosis of the eye or bruit auscultated.

Cranial magnetic resonance imaging and a standard catheter angiogram confirmed a left Barrow type D carotid cavernous fistula (CCF) supplied principally by the contralateral right external carotid artery (ECA) with involvement of the ipsilateral left internal carotid artery (ICA) intercavernous branches (Fig. 2A and B). There also appeared to be inferior petrosal sinus occlusion and bilateral cortical venous drainage.

Endovascular embolization via the inferior petrosal sinus was attempted twice, but the microcatheter could not be advanced due to sinus occlusion. Embolization was then attempted via the left superior ophthalmic vein, but the vein was too small to advance the microcatheter. A neurosurgical approach via craniotomy to the sphenoparietal sinus was then considered, but ultimately an ultrasound-guided left internal jugular vein retrograde approach was performed. A guide catheter was placed into the left jugular bulb and directed to the left inferior petrosal sinus and the posterior aspect of the cavernous sinus. Under fluoroscopic control, multiple coils were placed first in the most anterior aspect of the fistula and then in the inferior and posterior aspects of the cavernous sinus, respectively, to disconnect the fistula and the branches of the cortical venous drainage. Liquid Onyx was then instilled for complete obliteration of the CCF (Fig. 2C). The patient's abducens nerve (cranial nerve [CN] VI) palsy with ipsilateral Horner syndrome (Parkinson sign) persisted after the procedure.

Discussion

CCF is an abnormal connection between the ICA or ECA (or its branches) and the cavernous sinus (CS) that can be classified based on etiology (e.g., traumatic vs spontaneous), hemodynamics (e.g., high flow vs low flow), and anatomy (e.g., direct vs indirect). The Barrow classification system divides CCFs into 4 types: Type A fistulas are direct shunts between ICA and CS with high velocity blood flow, while types B–D are indirect dural shunts between the meningeal branches of the ICA or ECA and CS with low velocity blood flow.[1] Type B involves ICA branches, type C involves ECA branches, and type D involves both ICA and ECA branches.[1] Traumatic CCFs account for the majority of total CCFs and

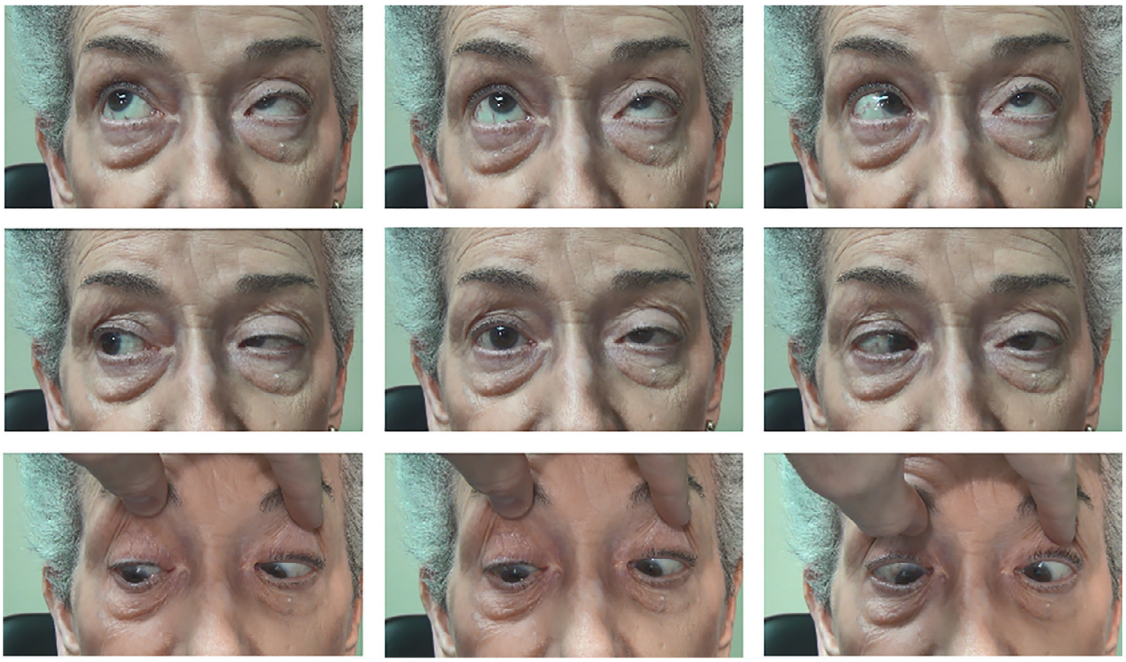

Fig. 1—Left abducens nerve palsy and ptosis. Esotropia and limitation of abduction in the OS consistent with abducens nerve palsy.

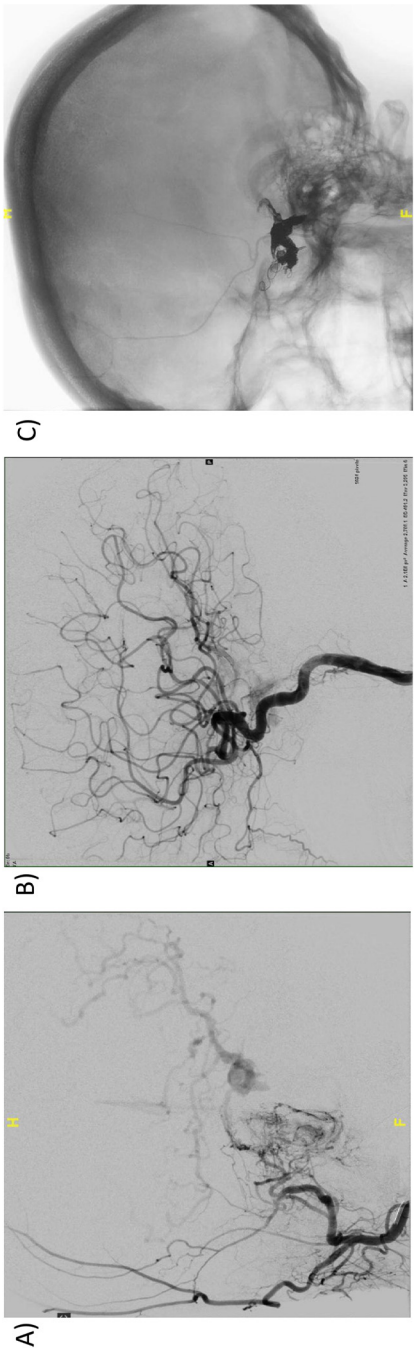

Fig. 2—Cerebral angiography of type D carotid cavernous fistula (CCF) before and after endovascular embolization. A, Right external carotid artery to left CCF with cortical venous drainage before embolization. B, Left internal carotid artery dural branches to the CCF before embolization. C, Occlusion of the CCF after embolization.

are almost always type A fistulas, while spontaneous CCFs tend to be types B—D.

Our patient had a type D CCF, which presented with a CN VI palsy with an ipsilateral Horner syndrome. This association was described by Parkinson in 1979[2] and localizes to the ipsilateral cavernous sinus. The CS structure contains CN III, CN IV, and the V1, V2 divisions of the trigeminal nerve in its lateral wall, while CN VI is embedded with substance of the sinus itself. In this location, the sympathetic nerve runs a short course close to CN VI on the ICA before it jumps onto the V1 segment of the trigeminal nerve. Although isolated CN VI palsy or Horner syndrome are nonlocalizing, the presence of both is a precise localizing sign that helps in narrowing down the location of the pathology and the diagnostic differentials based on other relevant patient demographics and history.[3] The ocular sympathetic nerve plexus in this location is the third order, postganglionic portion of a 3-neuron chain.

Parkinson sign has been reported with numerous different etiologies, including intracavernous carotid aneurysms,[4—6] metastatic tumours,[5,7,8] meningiomas,[9] and posttrauma CCFs.[10,11] To our knowledge however, this is the first case of Parkinson sign as the presenting finding of a type D CCF in the English language ophthalmic literature.

Supplementary Materials

Supplementary material associated with this article can be found in the online version at doi:10.1016/j.jcjo.2019.02.003.

Footnotes and Disclosure: The authors have no proprietary or commercial interest in any materials discussed in this article.

Iris Zhuang,[*] Ashwini Kini,[†] Bayan Al Othman,[†] Andrew G. Lee[†,‡,§,||,¶,,††]**

[*]Baylor College of Medicine, Houston, Texas; [†]Houston Methodist Hospital, Houston, Texas; [‡]MCPHS University School of Optometry, Worcester, Mass.; [§]Weill Cornell Medicine, New York, N.Y.; [||]University of Texas Medical Branch, Galveston, Texas; [¶]University of Texas MD Anderson Cancer Center, Houston, Texas; [**]Texas A & M College of Medicine, Bryan, Texas; [††]The University of Iowa Hospitals and Clinics, Iowa City, Iowa

Correspondence to:
Andrew G. Lee, MD; aglee@houstonmethodist.org

References

1. Barrow DL, Spector RH, Braun IF, Landman JA, Tindall SC, Tindall GT. Classification and treatment of spontaneous carotid-cavernous sinus fistulas. *J Neurosurg*. 1985;62:248–56.
2. Parkinson D, Bernard M. Horner syndrome and others? *Surg Neurol*. 1979;11:221–3.
3. Johnston J, Parkinson D. Intracranial sympathetic pathways associated with the sixth cranial nerve. *J Neurosurg*. 1974;40:236–43.

4. Abad JM, Alvarez F, Blazquez MG. An unrecognized neurological syndrome: sixth-nerve palsy and Horner's syndrome due to traumatic intracavernous carotid aneurysm. *Surg Neurol*. 1981;16:140–4.
5. Gutman I, Levartovski S, Goldhammer Y, Tadmor R, Findler G. Sixth nerve palsy and unilateral Horner's syndrome. *Ophthalmology*. 1986;93:913–6.
6. Striph GG, Burde RM. Abducens nerve palsy and Horner's syndrome revisited. *J Clin Neuroophthalmol*. 1988;8:13–7.
7. Tsuda H, Ishikawa H, Asayama K, Saito T, Endo S, Mizutani T. Abducens nerve palsy and Horner syndrome due to metastatic tumor in the cavernous sinus. *Intern Med*. 2005;44:644–6.
8. Tsuda H, Ishikawa H, Kishiro M, Koga N, Kashima Y. Abducens nerve palsy and postganglionic horner syndrome with or without severe headache. *Intern Med*. 2006;45:851–5.
9. Ebner RN, Ayerza DR, Aghetoni F. Sixth nerve palsy + ipsilateral Horner's Syndrome = Parkinson's Syndrome. *Saudi J Ophthalmol*. 2015;29:63–6.
10. Fujisawa H, Marukawa K, Kida S, Hasegawa M, Yamashita J, Matsui O. Abducens nerve palsy and ipsilateral horner syndrome: A predicting sign of intracranial carotid injury in a head trauma patient. *J Trauma*. 2001;50:554–6.
11. Kal A, Ercan ZE, Duman E, Arpaci E. Abducens nerve palsy and ipsilateral horner syndrome in a patient with carotid-cavernous fistula. *J Craniofac Surg*. 2015;26:e653–5.

Can J Ophthalmol 2018;54:e246–e249

https://doi.org/10.1016/j.jcjo.2019.02.003

Permanent functioning nasolacrimal fistula following extrusion of Jones tubes wrapped with conjunctival autograft

Excessive tearing caused by blocked tear ducts is a condition often encountered by the lacrimal surgeon. When the obstruction is due to canalicular damage, the most common management approach is conjunctivodacryocystorhinostomy (CDCR). This surgical technique, introduced by Jones in 1962,[1] consists of inserting a Pyrex® tube (Lester Jones tube; Gunther Weiss, Portland, OR) into the nasal cavity to bypass the blocked canaliculi completely.[1,2]

The success rate of Jones tube insertion has been variable, and complications can occur. Although the tube is well tolerated within the tissue, in some patients it can become displaced.[3] The most common complications include extrusion and malposition of the tube,[2,4–6] requiring tube replacement.[2–5]

Past attempts have been made to modify the Jones tube design so that the tube can remain in proper position. Flanged tubes have been tried as well as tubes made of frosted glass.[3,7] Recently, StopLoss Jones tubes (FCI Ophthalmic, Pembroke, MA) have been reported to have less extrusion rates, because of a silicone flange encircling the tube's medial end.[8,9]

Biological materials have been used for tear drainage in the past. Morax and Vialeix[10] first described using a skin graft to wrap a piece of paraffin wax placed into a newly created nasolacrimal passage. Vein grafts and buccal mucosal grafts were later used to wrap Jones tubes.[11,12] Campbell et al[12] wrapped their Jones tubes with buccal mucosa and reported 100% patency at 10–13 months, even after the Jones tube was removed from the tract.

We have trialled Jones tubes wrapped with conjunctival tissue, to improve surgical success rates of CDCR. Conjunctiva is easy to harvest and is the preferred graft source in pterygium surgery to prevent recurrence.[13,14] With its supply of lymphatic and blood vessels, autologous conjunctiva prevents scar tissue formation, which can cause the tube to contract and be displaced. A conjunctival wrap improves tube stability within the fistula tract, and results in less frequent tube replacements. Even if the tube extrudes, the conjunctival epithelialized tract remains patent and continues to allow for tear drainage.

A standard external dacryocystorhinostomy was performed before placement of a Jones tube. A tract was formed by using a 2.75 mm angled slit knife (Alcon, Mississauga, Canada) at the caruncle and aimed to enter the common canalicular opening. The 2.75 mm keratotome was used to widen the tract as needed to fit a 3.5-mm-diameter Jones tube. Conjunctiva was harvested from the superior limbal conjunctiva using fine Westcott scissors and 0.12 mm toothed forceps. Tenons capsule was carefully removed from the underside of the conjunctiva with the scissors. The graft's length was dependent on the length of the Jones tube. The graft's width was equal to $2\pi r$ ($r = 1.75$ mm for a 3.5-mm-diameter Jones tube; $2\pi r = 11.0$ mm). Conjunctiva is elastic, and so the actual dimensions of the graft were made slightly (1–2 mm) less than the calculated dimensions. Conjunctiva was then wrapped and sutured around the Jones tube using 6–0 Vicryl (Ethicon J555G, Markham, Canada) (Fig. 1). The epithelial side of the conjunctiva faced the Jones tube and the raw side faced outward. The tract for the Jones tube was splayed open with strabismus muscle hooks or any blunt, slender microinstrument of choice. The Jones tube was carefully lowered into position, taking care not to displace the conjunctival sleeve around it (Fig. 2). A double-armed 6–0 Prolene suture (Ethicon 8806H, Markham, Canada) was then wrapped around the neck of the Jones tube and tied through a rubber band bolster to secure the tube to the skin. This stay suture was removed after 7–10 days.

This technique has been used successfully in 2 patients requiring CDCR. Both had previous failed CDCRs. The average age of the patients at time of surgery was 59 years (range 54–64 years); both were female. One patient had her Jones tube inadvertently extruded during her sleep, 2 years after surgery. Her tract (without Jones tube) has now remained patent for 2.5 years. The second patient had her Jones tubes extruded after sneezing, 4 years after surgery. The fistula tract still remains patent 3.5 years after tube extrusion (Figs. 3 and 4). Both patients remain free of epiphora and discomfort, 4.5 years and 7.5 years after initial CDCR with conjunctival wrap, respectively.

The use of a conjunctival wrap around a Jones tube improves the success rate of tear drainage surgery, by keeping