CASE REPORT

Late-Onset Abducens Nerve Palsy after Endovascular Treatment for Carotid-Cavernous Fistula: Two Case Reports

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ABSTRACT

Endovascular treatment, including transarterial embolisation and transvenous embolisation, is generally considered effective for treating carotid-cavernous fistula. Immediate cranial nerve palsy following the procedure is the most common complication, but it usually resolves spontaneously. The authors report two cases of late-onset abducens nerve palsy after successful obliteration of direct carotid-cavernous fistulas, both of which required strabismus surgery.

Keywords: Abducens nerve palsy, carotid-cavernous fistula, transvenous embolisation

INTRODUCTION

Endovascular treatment, including transarterial embolisation (TAE) and transvenous embolisation (TVE), is generally considered effective for treating carotid-cavernous fistula (CCF). Immediate cranial nerve palsy (CNP) following the procedure is the most common complication, but it usually resolves spontaneously. We report two cases of late-onset abducens nerve palsy after successful obliteration of direct CCFs. Neuroimaging studies showed no evidence of CCF recanalisation. Both cases required strabismus surgery finally.

CASE REPORTS

Case I

A 50-year-old woman was diagnosed with left direct CCF with an initial presentation of conjunctival congestion and ocular hypertension in the left eye. Left abducens nerve paresis with diplopia was also

noted. She received TVE through left inferior petrosal sinus (IPS). A total of 14 Guglielmi and Matrix detachable coils were deployed at the cavernous sinus. Post-embolisation angiography showed total occlusion of the CCF without early venous drainage and the ICA was preserved. After a successful obliteration of the shunt, her ocular symptoms, including diplopia, improved significantly without any neurological sequelae. One month after treatment, new-onset oculomotor nerve palsy was detected in the left eye, but it completely resolved spontaneously in the following month. One year after treatment, gradual-onset left esotropia with mild lateral gaze limitation and diplopia were noted again, suggesting left abducens nerve palsy (Figure 1). Best corrected visual acuity was 20/20 (right eye) and 20/25 (left eye). Prism cover test revealed left esotropia of 30Δ, 25Δ on right gaze, and 35Δ on left gaze. There were no accompanying symptoms indicating recurrent CCF, such as conjunctival congestion, ocular hypertension, or tinnitus. Neuroimaging study was negative for recanalisation of the fistula. Diplopia was initially treated with prism glasses, but strabismus surgery,

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FIGURE 1 Clinical photographs of Case 1 demonstrating moderate abducens paresis in the left eye.

FIGURE 2 Clinical photographs of Case 2 demonstrating esotropia and limitation of abduction in the right eye.

which included left medial rectus muscle recession and lateral rectus muscle resection, was finally required. Her diplopia resolved after the surgery.

Case 2

A 12-year-old boy was diagnosed with right traumatic CCF after an accident causing closed head trauma. The initial presentation was conjunctival congestion of the right eye but there was no diplopia and gaze limitation. TAE was performed via right femoral artery. A total of 10 detachable coils were placed into the cavernous sinus, but residual fistula

remained. TVE via left femoral vein and inferior petrous sinus was performed then, a total of 9 coils were placed and 0.8cc liquid adhesive *N*-butyl-2-cyanoacrylate (N-BCA)/Lipiodol mixture was infused into cavernous sinus. However, the follow-up angiogram on the next day showed residual CCF so that another TVE through direct puncture of the ophthalmic vein was performed with Guglielmi detachable coils placed. The fistula was completely obliterated with preservation of blood flow in the ICA along with resolution of ocular symptoms and signs after the procedure. Six months later, however, gradual-onset right abducens nerve palsy with right esotropia and significant lateral gaze limitation were

FIGURE 3 Neuroimaging studies of Case 2. Before endovascular treatment, magnetic resonance angiography (MRA) (A) showed prominent CCF and engorged, arterialized right superior ophthalmic vein (SOV). T1-weighted coronal view of orbit (B) also showed engorged right SOV (arrow). Six months after the successful endovascular treatment of CCF, late-onset right abducens nerve palsy was noted in this patient. MRA (C) showed no residual direct or other indirect arteriovenous shunts at right cavernous sinus. T2-weighted coronal view (D) showed coil materials in right cavernous sinus. (arrow). No other abnormalities were found in brain parenchyma and ventricular system in this neuroimaging series.

noted (Figure 2). Best corrected visual acuity was 20/15 in both eyes. Prism cover test revealed right esotropia of 55Δ, more than 70Δ on right gaze, and 25Δ on left gaze. There were no other signs and symptoms suggesting recurrence of CCF. Neuroimaging studies showed no evidence of recanalisation of the fistula (Figure 3). Observation was suggested initially, but the abducens nerve palsy persisted without any improvement after a few months. The patient finally underwent strabismus

surgery including right medial rectus muscle recession and transposition of superior and inferior rectus muscles, which significantly improved the ocular alignment.

DISCUSSION

Carotid-cavernous fistula (CCF) is abnormal communication between the carotid arterial system and the

cavernous sinus. Symptoms are usually more significant when there are communications between the internal carotid artery and the cavernous sinus, which are traditionally called direct CCF. Endovascular treatment for symptomatic CCF is generally effective and infrequently associated with serious complications.^{1,2} The most common complication is CNP, which was previously reported to have an incidence rate of 10%–39%.^{3,4} The prognosis of CNP after endovascular treatment is favourable; spontaneous regression or improvement can be expected in a few months, with patients requiring only conservative treatment. Immediate aggravation of previous CNP or new-onset CNP after embolisation may be attributed to the mass effect of coil placement or direct injury of the nerve by the microcatheter. There may be a different mechanism for the onset of CNP a few days after the procedure. Progressive thrombosis or induced inflammation of the cavernous sinus may play a role in delayed cranial nerve dysfunction.³ In Case 1, transient oculomotor nerve palsy that occurred 1 month after TVE may be explained by this mechanism. However, the late, progressive onset of abducens nerve palsy that occurred more than 6 months after embolisation in both of our cases cannot be explained by the mechanisms mentioned above. Unlike most CNPs with a self-limiting course occurring after embolisation, late-onset CNPs in our cases had no sign of spontaneous regression after a period of conservative treatment, and strabismus surgery was finally required in both cases.

Recurrence of fistula may be a possible explanation for the late-onset abducens nerve palsy in our cases. The incidence of recanalisation after TVE was 9.1% according to a recent case series study.⁵ Rarely, recurrent CCF appearing at a new site posterior to the original fistula has also been shown in several case reports.⁶ However, in both of our cases, total obliteration of the fistula without a residual shunt was confirmed during the procedure; neither recurrent conjunctival congestion nor new-onset tinnitus suggesting recurrent fistula formation was noted. Evidence of recanalisation was not found in neuroimaging studies, either. Despite a negative finding in magnetic resonance angiography, an undetected subtle recanalisation could still exist, since this possibility could only be ruled out by further angiography. Another possible mechanism of nerve paresis could be due to direct compression by one or few of coils with subtle displacement. In both of our cases, multiple coils were placed in the cavernous sinus.

The abducens nerves run within cavernous sinus without dura matter protection is more sensitive and vulnerable. Unlike early CNPs following embolisation, which could be gradually improved by thrombus resolving, or as collateral circulation developed in cases with obstructed circulation of the vaso nervorum, late CNP caused by coil compression might not recover spontaneously.

In summary, we have presented two cases of late-onset abducens nerve palsy, which occurred months after a successful embolisation for CCF; both cases eventually required strabismus surgery. The underlying mechanism is unknown, but may be different from that of immediate or delayed CNPs after endovascular treatment, which are often self-limiting.

Declaration of interest: The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

Note: Figures 1 and 2 of this article are available in colour online at www.informahealthcare.com/oph.

REFERENCES

- [1] Meyers PM, Halbach VV, Dowd CF, Lempert TE, Malek AM, Phatouros CC, Lefler JE, Higashida RT. Dural carotid cavernous fistula: definitive endovascular management and long-term follow-up. *Am J Ophthalmol* 2002;134:85–92.
- [2] Gemmete JJ, Ansari SA, Gandhi DM. Endovascular techniques for treatment of carotid-cavernous fistula. *J Neuroophthalmol* 2009;29:62–71.
- [3] Nishino K, Ito Y, Hasegawa H, Kikuchi B, Shimbo J, Kitazawa K, Fujii Y. Cranial nerve palsy following transvenous embolization for a cavernous sinus dural arteriovenous fistula: association with the volume and location of detachable coils. *J Neurosurg* 2008;109:208–214.
- [4] Kim DJ, Kim DI, Suh SH, Kim J, Lee SK, Kim EY, Chung TS. Results of transvenous embolization of cavernous dural arteriovenous fistula: a single-center experience with emphasis on complications and management. *Am J Neuroradiol* 2006;27:2078–2082.
- [5] Yoshida K, Melake M, Oishi H, Yamamoto M, Arai H. Transvenous embolization of dural carotid cavernous fistulas: a series of 44 consecutive patients. *Am J Neuroradiol* 2010;31:651–655.
- [6] Hiu T, Horie N, Hayashi K, Kitagawa N, Morikawa M, Kawakubo J, Tsutsumi K, Suyama K, Nagata I. Recurrence of the cavernous sinus dural arteriovenous fistula at adjacent sinuses following repeated transvenous embolizations: case report and literature review. *Radiat Med* 2008; 26:431–437.