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A rare case of bilateral spontaneous indirect caroticocavernous fistula treated previously as a case of conjunctivitis

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Abstract

Carotid cavernous fistula is an abnormal communication between the carotid arterial system and the cavernous sinus. We present an interesting, rare case of bilateral spontaneous 'Barrow type- C' fistula treated presumptively as conjunctivitis. A 66 year old patient presented in the eye casualty at North Devon District Hospital in January 2016, referred from her General practitioner complaining of bilateral red eyes. She was found to have large, prominently diffused and engorged scleral blood vessels on both sides along with raised intraocular pressures of 26mm of Hg bilaterally. The patient was diagnosed with an indirect carotic cavernous fistulas bilaterally in view of the clinical and radiology findings. Barrow type - C dural fistulas were reported to be seen bilaterally on radiology findings. Patient was referred for interventional treatment to the closest neurosurgical center where she had four failed attempts of coil embolization after which she was referred to a second neurosurgery center at Bristol where she underwent successful coil catheterization as the treatment for her carotid cavernous fistula. Indirect carotid cavernous fistula most commonly occur spontaneously. Bilateral spontaneous indirect carotid cavernous fistula is a very rare diagnosis and and there are very few cases reported in the literature without an underlying etiology or a known cause like Ehlers -Danlos syndrome or diabetes mellitus. Bilateral spontaneous carotid cavernous fistulas are difficult to diagnose due to mild symptoms and no history of trauma. We conclude that carotid cavernous fistulas are a threat to the vision if left untreated due to delayed diagnosis. We recommend considering bilateral carotid cavernous fistula as a differential diagnosis in patients with an ongoing history of red eyes or those unresponsive to conventional topical treatment for conjunctivitis like symptoms.

Keywords: carotid artery aneurysm, carotid cavernous fistula, cavernous sinus, intraocular pressure

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Introduction

Carotid cavernous fistula (CCF) is an abnormal communication between the carotid arterial system and the cavernous sinus. CCF can be classified in a number of ways, as direct or indirect fistula based on anatomical features, traumatic or spontaneous on the basis of etiological features, or as high or low flow based on hematological basis. The most common etiology of direct fistula is a head trauma, as a result of which the internal carotid artery is damaged and a fistula is formed between the lacerated artery and the cavernous sinus. CCF uncommonly occurs spontaneously though. Barrow and colleagues[1] classified the CCF into direct (type A) and indirect (types B–D) types. Type A is high flow shunts between the internal carotid artery (cavernous portion) and the cavernous sinus and are usually caused by a trauma (rupture) of an internal carotid artery aneurysm.[1,2] Type A is more common in men. In women, older than 50 years, types B, C, and D are more common with 7:1 female to male ratio.[2,3] Types B, C, and D are fistulas between the cavernous sinus and extradural branches of the

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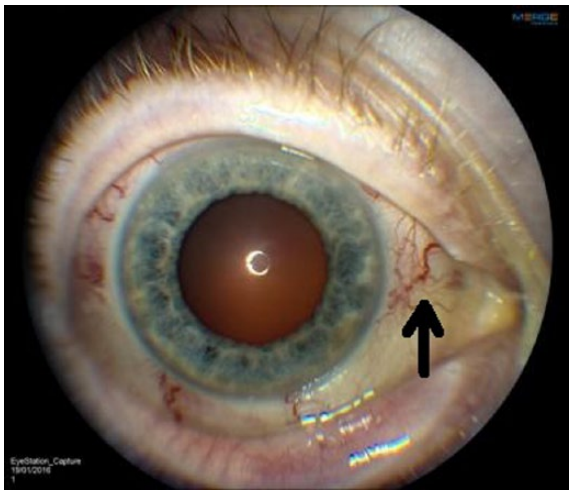

Figure 1. Visible engorged and dilated scleral blood vessels on the right eye (black arrow).

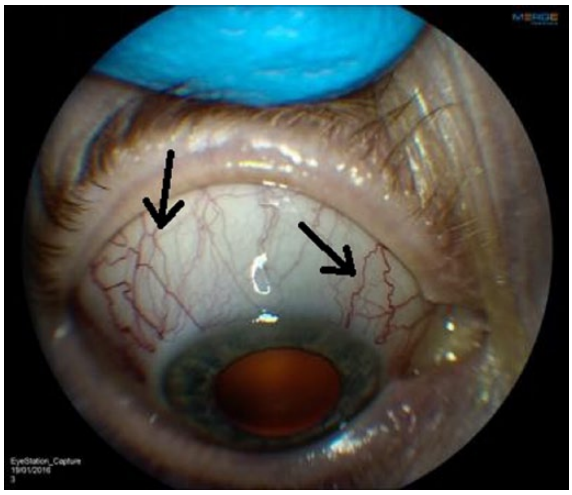

Figure 2. Superiorly visible engorged and dilated scleral blood vessels on the right eye (black arrows).

internal carotid artery, the external carotid artery, or both. A CCF is usually unilateral but less commonly bilateral and rarely bilateral and spontaneous in nature as observed in our case.

Case presentation

A 66-year-old patient presented in the eye casualty at North Devon District Hospital (Barnstaple, UK) in January 2016, referred from her general practitioner complaining of bilateral red eyes for about 3 weeks (Figures 1 and 2), diagnosed and treated presumptively as conjunctivitis, which was found to be nonresponsive to chloramphenicol 0.5% drops. On questioning, she had complaints of intermittent headache and a feeling of thumping in her head around the same time. She also described that at night she had been experiencing scratchy sounds in her ears for about the same duration of time. She had no complaints registered otherwise. There was no significant past ocular history. Medically, she was treated for hypothyroidism. Her medical history was unremarkable otherwise. There was no history of head or eye trauma.

On examination, the lady had an uncorrected visual acuity of 6/12 on the right eye and 6/6 on the left eye with no improvement of visual acuity on pinhole examination. There were large, prominently diffused, and engorged scleral blood vessels to be seen on both eyes. Visual fields were full to confrontation. Pupil examination was normal with normal reactivity on both eyes. There was no relative afferent pupillary defect. Her intraocular pressures (IOPs) were 19 mm Hg on the right eye and 20 mm Hg on the left eye. She was orthophoric in primary gaze, however, showed mild restriction of her extra-ocular movements bilaterally in horizontal gaze, suggesting both a mild abduction and adduction deficit in the two eyes along with slow saccadic movements overall. Neither proptosis nor bruit was observed at the time. No diplopia was reported by the patient. Fundus examination was unremarkable.

Diagnosis of spontaneous CCF, thyroid eye disease, and orbital varix were considered as differentials. Patient was seen again after 5 days and in review they had developed diplopia on looking at extreme gaze both right and left. She was found to have raised IOPs of 26 mm Hg bilaterally. A B-scan was performed which showed dilated superior ophthalmic veins bilaterally (Figures 3 and 4) which prompted an urgent magnetic resonance imaging (MRI) head scan to be carried out. Results showed dilated left superior ophthalmic vein along with enlarged cavernous sinus on the scan. Our colleagues from the radiology department suspected the same on right side but were not sure and hence the patient was referred by the local radiology department to a neuroradiology unit in a tertiary center for a further computed tomography angiography/MR angiography (CTA/MRA) scan. The CTA scan showed signs of bilateral CCF along with dilated superior ophthalmic veins on both sides (Figures 5 and 6). A further carotid angiogram confirmed bilateral CCF with a markedly dilated superior ophthalmic vein on the left side (Figure 7). The patient was diagnosed

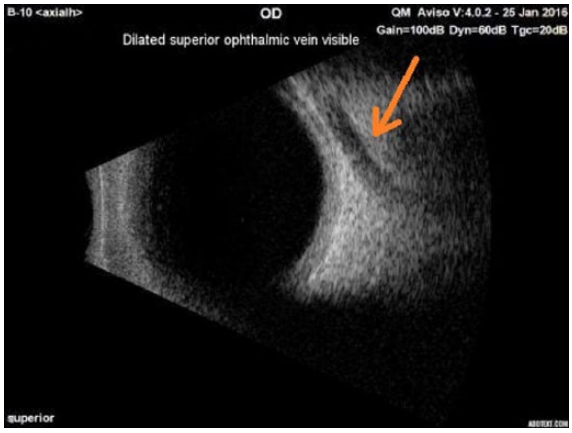

Figure 3. Dilated superior ophthalmic vein visible on the right eye scan (orange arrow).

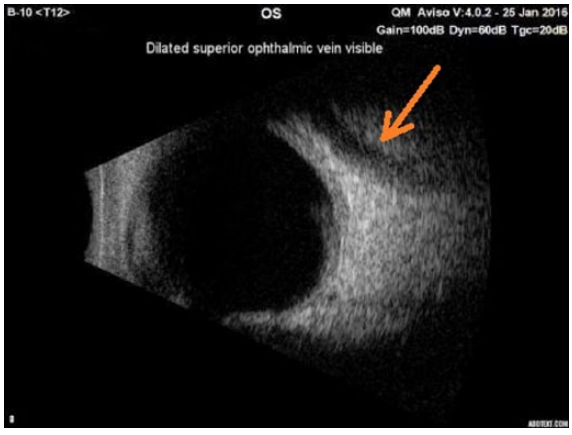

Figure 4. Dilated superior ophthalmic vein visible on the left eye scan (orange arrow).

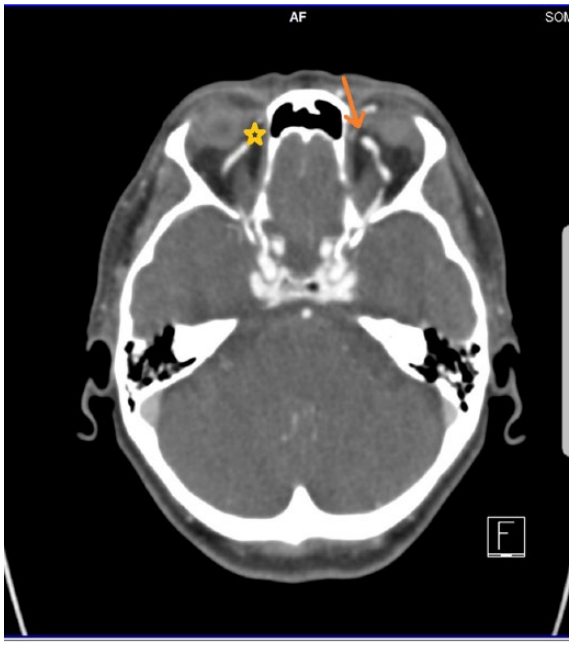

Figure 5. Computed tomography angiogram (CTA) image showing moderately dilated superior ophthalmic vein on the right side (yellow star) and a more marked dilated superior ophthalmic vein on the left side (orange arrow).

with an indirect CCF bilaterally in view of the clinical and radiology findings. Barrow type C-dural fistulas were reported to be seen bilaterally on the radiology findings; however, there were no other associated or underlying pathologies seen on the scans, and pathologies like orbital inflammatory disorders, space-occupying lesion, or cavernous sinus thrombosis were not observed on her MRI, CTA, or carotid angiogram. Patient in the mean time was reviewed again locally after 2 days in the clinic and was found to have a significant reduction in her vision with further increasing IOP. Her visual acuity was reduced to 3/60 bilaterally with IOP of 34 mm Hg on the right side and 32 mm Hg on the left side along with sluggish reaction of pupils on both sides; however, no anisocoria and no relative afferent pupillary defect were noted. She was started on Latanoprost eye drops to both eyes. The patient was also seen by an orthoptist for a formal Hess charting and was found to have a partial left-sided third nerve palsy with mildly reduced adduction on that side. A mild abduction deficit on both sides was also observed which was not clinical of sixth nerve palsy. Arrangements were made to refer the patient urgently to the closest neurosurgery center at Derriford Hospital (Plymouth, UK) where she was seen semi-urgently and four attempts were made to embolize the fistula with coil catheter. However, all four attempts failed and she was referred to a second neurosurgical center in Bristol where she underwent coil catheterization successfully as the treatment for her CCF. After 2 months of the interventional procedure, her symptoms were resolved and her IOP was back to normal at around 16 mm Hg in both eyes along with restoration of full extra-ocular movements on both sides and a normal vision of 6/9 bilaterally.

Discussion

CCF is found more commonly as a result of trauma; however, a spontaneous CCF is not uncommon. Helmke and colleagues[4] in their study showed 42 cases of type A nontraumatic CCF. They further suggested that a sudden increase in the intraluminal

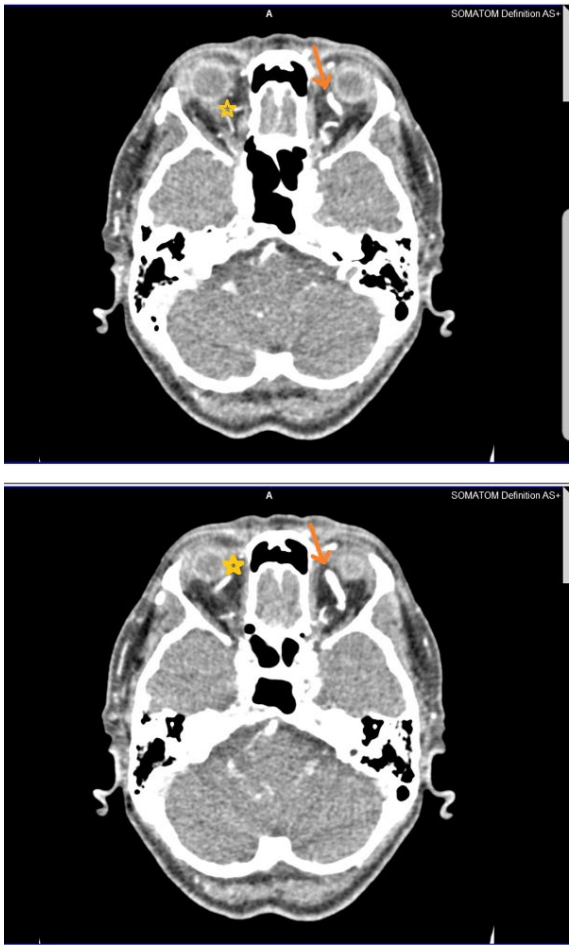

Figure 6. Computed tomography angiogram (CTA) images showing prominent superior ophthalmic veins on the right side (yellow stars) and tortuously dilated superior ophthalmic veins on the left side (orange arrows).

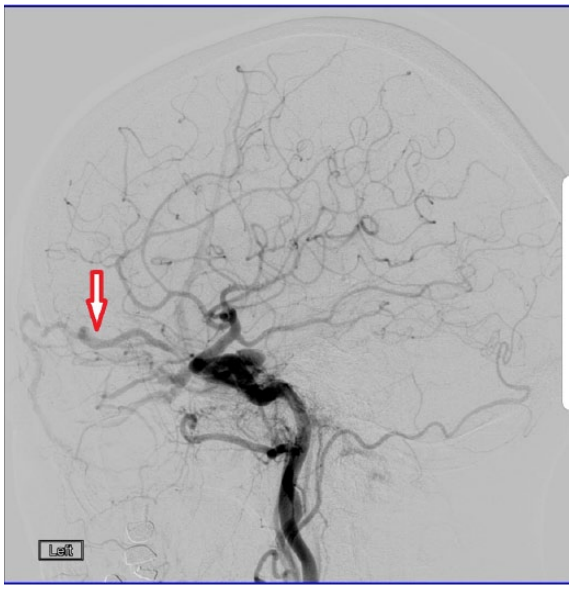

Figure 7. Carotid angiogram lateral view showing CCF with a dilated left side superior ophthalmic vein (arrow).

pressure ruptured the internal carotid artery which may explain the nontraumatic cause of these. Diagnosis of CCFs should be considered with bilateral eye symptoms and bilateral nerve palsies.[5] Treatment of CCF is mainly interventional in nature; however, it includes observation and medical management in a few cases. Type A fistulas very rarely resolve spontaneously; however, type B, C, and D fistulas have a higher incidence of spontaneous resolution.[6,7] Therefore, some cases of indirect and low-flow fistulas can initially be observed only or managed conservatively for ocular symptoms with medical management or manual carotid compression.[8] Interventional treatment options include both surgical and endovascular options which are ligation of the external or internal carotid arteries, fistula embolization with glue, microcoils, and stents and detachable ballons.[9]

In summary, indirect CCF most commonly occurs spontaneously.[10] Bilateral spontaneous indirect CCF is a very rare diagnosis and there are very few cases reported in the literature without an underlying etiology or a known cause like Ehlers–Danlos syndrome or diabetes mellitus. A recent case report and review article have mentioned only 26 reported cases in literature excluding their 2 cases of bilateral nontraumatic spontaneous CCFs according to their review of literature since 1963.[11] Our literature search has found a total of 35 reported cases of spontaneous, nontraumatic bilateral CCFs since 1963 now including our case. We found another six cases in our review of literature that were not included in this recent literature review along with the two additional cases which they have reported in their article.[11] Table 1 shows a summary of all reported 35 cases of bilateral spontaneous CCF with patient demographic data, reported presentation, treatment, and outcomes. There have been only two reported cases[12,13] of bilateral type C fistulas in the literature out of the 35 reported cases of bilateral CCFs (Table 1) which make this case report very rare as our case is presented as the only third case of bilateral Barrow type C fistulas in the literature. Bilateral spontaneous CCFs are difficult to diagnose due to mild symptoms and no history of trauma. B-scan ultrasound is an easy investigative tool which shows dilated superior

Table 1. Review of the bilateral spontaneous carotid cavernous fistula (CCF) reported cases.

No.	References	Age at presentation and sex	Initial presentation	Type of CCF	Treatment	Outcome
1	Jedrzejowska and colleagues[20] (Polish)	N/A	Unknown	R-type B L-type B	Unknown	Unknown
2	Schoolman and Kepes[21]	39, F	Scleral injection, decreased visual acuity, and protrusion of left eye with diplopia	R-Unknown L-Unknown	Bilateral surgical ligation	Death from pericardial hemorrhage
3	Voigt and colleagues[22]	53, F	Intracranial murmur with right-sided proptosis and sixth nerve palsy	R-Type A L-Type A	Conservative management	CCFs resolved, symptoms fully resolved
4	Taptas[23] (French)	45, F	Unknown	R-Unknown L-Unknown	Surgical embolization	Unknown
5	Stolpmann[24] (German)	66, F	Unknown	R-Unknown L-Unknown	Conservative, carotid compression	Unknown
6	Manaka and colleagues[25] (Japanese)	N/A	Unknown	R-Unknown L-Unknown	Unknown	Unknown
7	Rainer and Haselbach[26] (German)	61, F	Unknown	R-Unknown L-Unknown	Conservative	Unknown
8	Kato and colleagues[12] (Japanese)	52, F	Left side severe headache, weakness of the left extra-ocular muscles and left ptosis	R-Type C L-Type C	Conservative	Unknown
9	Kato and colleagues[12] (Japanese)	50, M	Right ptosis, headache, and diplopia	R-Unknown L-Unknown	Unknown	Unknown
10	Kato and colleagues[12] (Japanese)	U/K	Unknown	R-Unknown L-Unknown	Unknown	Unknown
11	Oishi and colleagues[13] (French)	55, F	Bilateral ophthalmoplegia, bilateral chemosis, conjunctival injection	R-Type C L-Type C	Unknown	Unknown
12	Diez Lobato and colleagues[27]	68, F	Exophthalmos and injection of the conjunctiva on the left side	R-Type C L-Type B	Conservative	Unresolved CCFs, refused treatment
13	Desai and colleagues[28]	38, F	Headache, dimness of vision and exophthalmos in right eye, bruit	R-Type A L-Type A	Balloon embolization	Resolved CCFs, partial recovery of symptoms
14	vd Vliet and colleagues[29]	70, F	Swelling of right eyelid and redness of right eye with pulsating whizzing sound, proptosis	R-Type A L-Type A	Conservative	Spontaneously regressed

(Continued)

Table 1. (Continued)

No.	References	Age at presentation and sex	Initial presentation	Type of CCF	Treatment	Outcome
15	Labbe and colleagues[30] (French)	U/K	Increased intraocular pressure	R-Unknown L-Unknown	Transvenous coil embolization, sclerotherapy	Unknown
16	Courtheoux and colleagues[31]	60, F	Bilateral conjunctival injection, mild exophthalmos, chemosis, and increased intraocular pressure	R-Type C L-Type B	Bilateral, staged-coil embolization and sclerotherapy	CCF resolved, symptoms resolved
17	Albert and colleagues[32]	64, F	Bilateral exophthalmos, conjunctival hyperemia with marked chemosis, left abducens palsy, and bilateral engorgement of the optic disc	R-Type D L-Type B	Staged, bilateral-surgical arterial embolization	CCF resolved, symptomatically improved
18	Haugen and colleagues[33]	74, M	Left sided exopthlamos, chemosis, and dilated episcleral veins	R-Type B L-Type B	Conservative	Spontaneous resolution of fistulas with symptomatic improvement
19	Chaloupka and colleagues[34]	40, F	Proptosis, chemosis, and conjunctival injection of the right eye; partial third and sixth cranial nerve palsy on the right	R-Type D L-Type D	Unilateral transvenous embolization	CCF resolved
20	Berlis and colleagues[35]	74, F	Diplopia, exophthalmos on the left side, scotomas, left visual blur, and left conjunctival injection	R-Type D L-Type D	Bilateral transvenous coil embolization	Resolved CCFs, complete recovery
21	Jethani and Ajani[36]	53, M	Bilateral chemosis and redness, restriction of movement in all directions of gaze, best-corrected vision 5/60 in his right eye and 6/36 in his left eye	R-Type D L-Type D	Ophthalmic surgery	Visual acuity improved to 6/24 unaided
22	Dabus and colleagues[37]	69, F	Progressive double vision due to left sixth nerve palsy, pulsatile tinnitus, bilateral eye pain, and intense bilateral conjunctival chemosis	R-Type D L-Type D	Unilateral transvenous coil embolization	Resolved CCFs, transient worsening but symptoms resolved
23	Wong and colleagues[38]	74, F	2-month history of diplopia, blurring of vision and left eye pain; left proptosis, left eye chemosis and left abducens nerve palsy	R-Type B L-Type C	Transvenous coil embolization	Resolved CCFs, symptoms resolved
24	Girardin and colleagues[39] (French)	34, F	Unknown	R-Unknown L-Unknown	Unknown	Unknown
25	Amorim and colleagues[40]	36, M	Headaches, diplopia, and blurry vision, sixth nerve palsies bilaterally, impaired visual acuity	R-Type D L-Type D	Transvenous coil embolization	CCFs untreated, symptoms resolved
26	Bilbin-Bukowska and colleagues[41] (Polish)		Unknown	R-Unknown L-Unknown	Unknown	Unknown

Table 1. (Continued)

No.	References	Age at presentation and sex	Initial presentation	Type of CCF	Treatment	Outcome
27	Dowlut and colleagues[42]	78, F	1-week history of horizontal diplopia secondary to left sixth nerve palsy, bilateral corkscrew episcleral vessels, pulsatile elevated IOPs	R-Unknown L-Unknown	Coil embolization	CCF resolved with resolved symptoms and visual acuity of 6/12 in the right eye and 6/9 in the left eye
28	Kwon and colleagues[43]	46, F	2-month history of headache, diplopia, bilateral exophthalmos, and conjunctival injection	R-Type D L-Type D	Transvenous coil embolization initially then coil embolization via superior ophthalmic vein route by direct surgical exposure	CCG resolved 2 months after last embolization and symptoms resolved
29	Liberatore and Lechan[44]	53, F	Left sixth nerve palsy and enlarged pituitary on MRI head. Headaches sinus pressure and bilateral eye redness 3 months prior to admission	R-Unknown L-Unknown	Bilateral endovascular coiling	CCF resolved, symptoms resolved
30	Jun and colleagues[45] (Korean)	53, F	Progressive bilateral chemosis, exophthalmos and sixth nerve palsy on admission and history of painful ophthalmoplegia since 8 months	R-Unknown L-Unknown	Multiple attempts of transarterial, transvenous embolization with gelform material and platinum coils	Partial resolution of symptoms
31	Jun and colleagues[45] (Korean)	45, F	Slowly progressive headache, ptosis, left pupil dilation, and diplopia suggesting left inferior rectus paralysis	R-Unknown L-Unknown	Unknown	Unknown
32	Al-Mufti and colleagues[11]	57, M	Progressive worsening left eye pain, bilateral chemosis, proptosis, and periorbital swelling and history of 2 weeks prior double vision with bilateral loss of visual acuity	R-Type D L-Type D	Transvenous embolization of left cavernous sinus and inter cavernous sinus	CCFs resolved with immediate resolution of symptoms
33	Al-Mufti and colleagues[11]	77, M	3-week history of diplopia, blurry vision, and right eyelid droop that had recently worsened to right-sided chemosis, proptosis, and exophthalmos	R-Type D L-Type B	Conservative management	Complete resolutions of CCFs and symptoms at 4 months
34	Belhachmi A[46]	22, F	History of chronic headaches and progressive bilateral exophthalmitis of both eyes since 4 months	R-Unknown L-Unknown	Embolization with releasable balloons	Complete resolution of both CCFs with symptoms
35	Our study	66, F	History of bilateral red eyes for 3 weeks treated as conjunctivitis. Intermittent headache and a feeling of thumping in her head around the same time along with scratchy sounds in both ears	R-Type C L-Type C	Transvenous coil embolization of fistulas	CCFs resolved bilaterally. Symptoms completely resolved

ophthalmic veins in CCF patients;[14–16] hence, CCF can mostly be diagnosed easily in the smaller community hospitals or large general practices with it if the access to B-scan is available. In difficult cases where further investigation is required, MRI head scan becomes necessary as it is critical in diagnosing CCF. Diagnosis may be delayed due to not having the access to MRI facility locally; hence, an important aspect of early diagnosis in some cases is dependent on the ease of access to MRI facility. Optical coherence tomography angiography (OCT-A) is a noninvasive investigation previously limited to retina and posterior segment examinations but is now beginning to be used for assessment of anterior segment vasculature and may hold some utility in evaluation of patients with suspected CCF.[17,18] A case report by some researchers have used OCT-A of the anterior segment for studying the delineation of abnormal episcleral venous plexus secondary to dural CCF.[18] We conclude that CCFs are a threat to the vision if left untreated due to delayed diagnosis. Raised venous pressure and IOP in a CCF patient may compromise the retinal perfusion and result in loss of visual acuity.[19] Also, raised IOP as seen in our case can cause damage to the optic nerve by causing secondary glaucoma due to persistently raised IOP if CCF is not treated or there is a delay in the treatment. We recommend considering bilateral CCF as a differential diagnosis in patients with an ongoing history of red eyes or those unresponsive to conventional topical treatment for conjunctivitis-like symptoms.

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Conflict of interest statement

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Informed consent

A written informed consent was obtained from the patient for this case report information and for the images related to the case report to be published anonymously for educational and research purposes.

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References

1. Barrow DL, Spector RH, Braun IF, *et al.* Classification and treatment of spontaneous carotid-cavernous sinus fistulas. *J Neurosurg* 1985; 62: 248–256.
2. Kurata A, Takano M, Tokiwa K, *et al.* Spontaneous carotid cavernous fistula presenting only with cranial nerve palsies. *Am J Neuroradiol* 1993; 14: 1097–1101.
3. Lewis AI, Tomsick TA, Tew JM, *et al.* Management of 100 consecutive direct carotid-cavernous fistulas: results of treatment with detachable balloons. *Neurosurgery* 1995; 36: 239–245.
4. Helmke K, Krüger O, Laas R, *et al.* The direct carotid cavernous fistula: a clinical, pathoanatomical, and physical study. *Acta Neurochir* 1994; 127: 1–5.
5. Karadag R, Bayraktar N, Kirbas I, *et al.* Unilateral, indirect spontaneous caroticocavernous fistula with bilateral abduction palsy. *Indian J Ophthalmol* 2011; 59: 336–337.
6. Murthy TVSP, Chandra M, Gupta P, *et al.* Caroticocavernous fistula—a case report. *Indian J Anaesth* 2005; 49: 220–222.
7. Naragum V, Barest G, Abdalkader M, *et al.* Spontaneous resolution of post-traumatic direct carotid-cavernous fistula. *Intervent Neurol* 2018; 7: 1–5.
8. Gemmete JJ, Chaudhary N, Pandey A, *et al.* Treatment of carotid cavernous fistulas. *Curr Treat Options Neurol* 2010; 12: 43–53.
9. Korkmazer B, Kocak Tureci E, Islak C, *et al.* Endovascular treatment of carotid cavernous sinus fistula: a systematic review. *World J Radiol* 2013; 5: 143–155.
10. Stetler WR Jr, Chaudhary N, Wilson TJ, *et al.* Indirect carotid-cavernous fistula following minor head trauma treated with incomplete radiographic endovascular occlusion. *BMJ Case Rep* 2012; 2012: bcr0320126004.
11. Al-Mufti F, Amuluru K, El-Ghanem M, *et al.* Spontaneous bilateral carotid-cavernous fistulas secondary to cavernous sinus thrombosis. *Neurosurgery* 2017; 80: 646–654.
12. Kato M, Maki Y, Nakada Y, *et al.* Three cases of spontaneous bilateral external carotid-cavernous sinus fistula (dural arteriovenous shunts in the region of the cavernous sinus) (author's transl.). *No Shinkei Geka* 1975; 3: 607–613.
13. Oishi M, Nakahara K, Fukuuchi Y, *et al.* A case of bilateral spontaneous external carotid-cavernous fistulae with intrahepatic gallbladder

(author's transl.). *Rev Neurol* 1979; 135: 245–250.

14. Jorgenson JS and Guhoff R. Differential diagnosis of the dilated superior ophthalmic vein by B-scan ultrasonography. *Orbit* 1986; 5: 259–262.
15. Nemeth J, Nagy E, Morvay Z, *et al.* Diagnosis of carotid-cavernous sinus fistula using ultrasound, color Doppler imaging, CT-scan and digital subtraction angiography. In: Tane S, Thijssen JM and Fledelius HC (eds) *Ultrasonography in ophthalmology*. Dordrecht: Springer, 1995, pp. 211–215.
16. Spector RH. Echographic diagnosis of dural carotid-cavernous sinus fistulas. *Am J Ophthalmol* 1991; 111: 77–83.
17. Ang M, Sim DA, Keane PA, *et al.* Optical coherence tomography angiography for anterior segment vasculature imaging. *Ophthalmology* 2015; 122: 1740–1747.
18. Ang M, Sng C and Milea D. Optical coherence tomography angiography in dural carotid-cavernous sinus fistula. *BMC Ophthalmol* 2016; 16: 93.
19. Connors JJ and Wojak JC. *Interventional neuroradiology: strategies and practical techniques*. Philadelphia, PA: W. B. Saunders Co., 1999, pp. 215–226.
20. Jedrzejowska H, Mossakowski M and Rajszys R. Spontaneous bilateral arteriovenous fistula between the internal carotid artery and the cavernous sinus. *Pol Tyg Lek* 1963; 18: 1153–1158.
21. Schoolman A and Kepes JJ. Bilateral spontaneous carotid-cavernous fistula in Ehlers-Danlos syndrome: case report. *J Neurosurg* 1967; 26: 82–86.
22. Voigt K, Sauer M and Dichgans J. Spontaneous occlusion of a bilateral caroticocavernous fistula studied by serial angiography. *Neuroradiology* 1971; 2: 207–211.
23. Taptas JN. The treatment of carotid-cavernous aneurysms with embolization of the cervical internal carotid. Apropos of 3 cases, including a case of bilateral carotid-cavernous aneurysm treated with muscular embolization. *Rev Neurol* 1971; 124: 277–290.
24. Stolpmann E. Bilateral, nontraumatic carotid artery-cavernous sinus fistula. *Albrecht Von Graefes Arch Klin Exp Ophthalmol* 1972; 185: 83–94.
25. Manaka S, Fukushima T, Hori T, *et al.* Case of spontaneous bilateral external carotid-cavernopetrosal sinus fistula. *No To Shinkei* 1971; 23: 71–77.
26. Rainer A and Haselbach H. Spontaneous bilateral carotid-cavernous-fistula (author's transl.). *Laryngol Rhinol Otol* 1975; 54: 163–168.
27. Diez Lobato R, Escudero L and Lamas E. Bilateral dural arteriovenous fistula in the region of the cavernous sinus. *Neuroradiology* 1978; 15: 39–43.
28. Desai MT, Iyengar PS, Oza MK, *et al.* Bilateral carotid-cavernous fistulae—a case report. *J Assoc Physicians India* 1986; 34: 520–522.
29. vd Vliet AM, Rwiza HT, Thijssen HO, *et al.* Bilateral direct carotid-cavernous fistulas of traumatic and spontaneous origin: two case reports. *Neuroradiology* 1987; 29: 565–569.
30. Labbe D, Courtheoux P, Rigot-Jolivet M, *et al.* Bilateral dural carotid-cavernous fistula. Its treatment by way of the superior ophthalmic vein. *Rev Stomatol Chir Maxillofac* 1987; 88: 120–124.
31. Courtheoux P, Labbe D, Hamel C, *et al.* Treatment of bilateral spontaneous dural carotid-cavernous fistulas by coils and sclerotherapy. Case report. *J Neurosurg* 1987; 66: 468–470.
32. Albert P, Polaina M, Trujillo F, *et al.* Direct carotid sinus approach to treatment of bilateral carotid-cavernous fistulas. Case report. *J Neurosurg* 1988; 69: 942–944.
33. Haugen OH, Sletteberg O, Thomassen L, *et al.* Bilateral non-traumatic carotid cavernous sinus fistula with spontaneous closure. *Acta Ophthalmol* 1990; 68: 743–747.
34. Chaloupka JC, Goller D, Goldberg RA, *et al.* True anatomical compartmentalization of the cavernous sinus in a patient with bilateral cavernous dural arteriovenous fistulae. Case report. *J Neurosurg* 1993; 79: 592–595.
35. Berlis A, Klisch J, Spetzger U, *et al.* Carotid cavernous fistula: embolization via a bilateral superior ophthalmic vein approach. *Am J Neuroradiol* 2002; 23: 1736–1738.
36. Jethani J and Ajani JK. Cataract extraction in spontaneous low-flow indirect dural bilateral carotid cavernous fistula. *Graefes Arch Clin Exp Ophthalmol* 2006; 244: 404–406.
37. Dabus G, Batjer HH, Hurley MC, *et al.* Endovascular treatment of a bilateral dural carotid-cavernous fistula using an unusual unilateral approach through the basilar plexus. *World Neurosurg* 2012; 77: 201.e5–201.e8.
38. Wong GK, Sze AM, Yu SC, *et al.* Diffuse large B-cell non-Hodgkin's lymphoma associated with bilateral carotid-cavernous fistulas in an elderly woman. *J Clin Neurosci* 2007; 14: 904–907.

39. Girardin M, Puzenat E, Humbert P, *et al.* Bilateral spontaneous carotid-cavernous fistula revealing Ehler-Danlos disease. *Ann Dermatol Venereol* 2013; 140: 296–299.

40. Amorim E, Jankowitz BT, Jovin TG, *et al.* Recovery from ophthalmoplegia and proptosis after repair of bilateral carotid-cavernous sinus fistulas. *JAMA Neurol* 2013; 70: 1584–1585.

41. Bilbin-Bukowska A, Stepien A, Brzozowski K, *et al*. Diagnostic and therapeutic problems of bilateral carotid-cavernous sinus fistula. *Pol Merkur Lekarski* 2014; 36: 345–347.

42. Dowlut M, Quinlan M and Barry JS. Cataract surgery and preexisting bilateral carotid cavernous fistula. *JCRS Online Case Rep* 2013; 1: e17–e18.

43. Kwon SC, Kwon Y, Kim JM, *et al*. Endovascular treatment of bilateral spontaneous carotid-cavernous fistulas. *J Korean Neurosurg Soc* 2004; 36: 150–152.

44. Liberatore A and Lechan RM. Bilateral carotid-cavernous fistulas: an uncommon cause of pituitary enlargement and hypopituitarism. *Case Rep Endocrinol* 2016; 2016: 6364203.

45. Jun SM, Park KW, Gha JK, *et al.* Bilateral spontaneous carotid-cavernous fistula (CCF) presenting as painful ophthalmoplegia: two cases. *J Korean Neurol Assoc* 1997; 15: 1148–1154.

46. Belhachmi A. Spontaneous bilateral carotid-cavernous fistulas: about a case and review of the literature. *Pan Afr Med J* 2017; 27: 91.