Dural Carotid–Cavernous Sinus Fistula and Central Retinal Vein Occlusion: A Case Report and a Review of the Literature

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A case of dural carotid–cavernous sinus fistula was complicated by hemorrhagic retinopathy due to central retinal vein occlusion. A 57-year-old woman with a dural carotid–cavernous sinus fistula was initially treated by transarterial particulate embolization. Her symptoms subsequently improved, but 4 months later she gradually developed decreased visual acuity due to central retinal vein occlusion. A review of the literature showed that central retinal vein occlusion may be more common than previously thought. Dural carotid–cavernous sinus fistulas should be treated and followed taking into account possible complication by central retinal vein occlusion. Early detection of central retinal vein occlusion by frequent ophthalmologic examinations may prevent deterioration of visual acuity.

KEY WORDS: Carotid–cavernous sinus fistula; Central retinal vein occlusion; Embolization; Retinal hemorrhage

A dural carotid–cavernous sinus fistula (CCF) involves the communication of the dural branches of the internal and/or external carotid arteries with the cavernous sinus. Clinical symptoms include conjunctival injection, chemosis, proptosis, extraocular nerve palsy, retroorbital pain, and subjective/objective bruits [9]. The condition commonly occurs in middle-aged women and has a generally favorable prognosis.

Central retinal vein occlusion (CRVO) is considered to be a rare complication of this disease [1,4,10–12,16,18,20] (Table 1). In the case reported here, clinical improvement was initially obtained by transarterial embolization of the branches of the external carotid artery using polyvinyl alcohol particles. Four months after embolization, however, the patient developed CRVO with poor ocular outcome.

This case report also offers a review of the literature and presents a possible interpretation of this condition.

Case Report

This 57-year-old woman experienced a left parietal headache in May 1989, which subsequently moved to the left retroorbital area. The following month she noticed diplopia with subjective bruit, which was synchronous with her heart beats. She had no notable past or family history of head trauma, hypertension, diabetes mellitus, or glaucoma.

She was first examined in another clinic in June 1989. At that time she had mild left abducent palsy and modest proptosis, conjunctival injection, and objective bruits over the left eye. Her corrected visual acuity was 20/20 in the right eye and 20/20 in the left. The optic fundi were normal bilaterally. Cerebral angiography demonstrated a dural CCF on the left side. The feeding arteries were the bilateral ascending pharyngeal arteries and the left middle meningeal artery in the external carotid system, and the bilateral meningohypophyseal trunks of the internal carotid system. Drainage from the left cavernous sinus was to the left superior ophthalmic vein and to the right inferior petrosal vein through the intercavernous vein (Figure 1).

The patient was then referred to our clinic in early July 1989. Her corrected visual acuity on admission was 20/17 in the right eye and 20/20 in the left, the intraocular pressure was 15 mmHg in the right eye and 38 mmHg in the left, and proptosis was 8 mm in the right eye and 12 mm in the left. Thereafter acetazolamide and a beta-blocker administered through an eyedropper, were used for control of her intraocular pressure. On July 5, 1989, transvenous embolization of the left cavernous sinus was attempted through the right inferior petrosal sinus and

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Table 1. *Summary of the Dural Carotid–Cavernous Sinus Fistulas with Central Retinal Vein Occlusion*

Author	Age	Sex	CRVO occurred after embolization	Mode of therapy	Visual acuity
Nishimura et al [10]	36	F	No	C	Good
	54	F	No	C	Good
	62	F	No	C	Good
	59	F	No	C	Good
Terubayashi et al [18]	65	F	No	C	Good
Nukui et al [11]	64	F	No	C	Poor
	72	F	No	C	Poor
Pollock and Miller [12]	68	F	No	C	Poor
Brunette and Boghen [1]	47	F	No	C	Poor
Yoshida et al [20]	54	F	Yes	A	Good
	64	F	Yes	A	Good
	59	M	Yes	A	Good
Suzuki et al [16]	46	F	No	A	Poor
Hashimoto et al [4]	21	M	Yes	A + V	Poor
Present report (1990)	57	F	Yes	A	Poor

Abbreviations: A, transarterial embolization; C, conservative therapy; CRVO, central retinal vein occlusion; F, female; M, male; V, transvenous embolization.

the intercavernous sinus. However, venous drainage had already changed solely to the left superior ophthalmic vein, so transarterial embolization of the left ascending pharyngeal artery and the left middle meningeal artery, using polyvinyl alcohol particles sized 250 to 600 μm, was carried out (Figure 2).

This embolization resulted in clinical improvement of the patient's chemosis, proptosis, and abducent palsy. Her corrected visual acuity remained at 20/17 in the right eye, and it improved at 20/13 in the left. Her intraocular pressure stayed at 15 mmHg in the right eye and decreased to 23 mmHg in the left. A scattered dot retinal hemorrhage was also noted in the left eye. Although her abducent palsy and chemosis further improved, and bruit was no longer audible in October, she complained of a gradual decrease in left visual acuity in November 1989.

Early in December, her corrected visual acuity was 20/13 in the right eye and 20/130 (NC) in the left, with an intraocular pressure of 12 mmHg in the right eye and 19 mmHg in the left. A marked retinal hemorrhage and CRVO were noted on the left side, and cystoid macula edema was also eminent. Cerebral angiography revealed minimum shunted flow in the left cavernous sinus and in the left superior ophthalmic vein, fed solely by the bilateral meningohypophyseal trunks of the internal carotid arteries (Figure 3). She was followed conservatively. At the final follow-up in January 1990, her visual acuity was 20/17 in the right eye and 20/100 (NC) in the left.

Discussion

A CRVO represents two clinical conditions: one is simple occlusion of the central retinal vein, which is referred to as a "venous stasis retinopathy," for which a good prognosis can be expected; the other is occlusion of the central retinal vein with retinal ischemia, which is referred to as "hemorrhagic retinopathy," which has a usually poor prognosis [6]. Retinal hemorrhage has been reported as a complication of dural CCF [8,15]. Although the cause of retinal hemorrhaging seems to be CRVO, the exact etiology has not been clearly documented. Dural CCFs associated with CRVO have been reported so far in 15 cases, including our own case [1,4,10–12,16,18,20]. Among these 15 cases, 8 cases had good visual acuity, while in the remaining 7 cases visual acuity was poor. There were 13 women and 2 men, ranging in age from 21 to 72 years, with a mean age of 55 years. The predominance of females in these 15 cases is consistent with the findings for dural CCFs.

Anatomically, the central retinal vein connects with the superior ophthalmic vein, which connects with the cavernous sinus. Seeger et al [13] observed thrombosis formation in the superior ophthalmic vein and the cavernous sinus in dural CCFs. Sergott et al [14] reported the transient deterioration of the ocular symptoms as "paradoxical worsening" due to thrombosis formation in the superior ophthalmic vein. It is easily understandable that sinus thrombosis in the cavernous sinus may extend into the superior ophthalmic vein, producing CRVO. However, CRVO itself does not cause any visual damage, probably due to anastomosis between the central retinal vein and the choroidal circulation of the eye [5]. When the thrombosis involves these anastomotic veins, however, it produces retinal ischemia and hemorrhagic retinopathy. "Paradoxical worsening" due to thrombosis formation is reported to have a good ocular prognosis and conservative follow-up is recommended [14], but it may actually

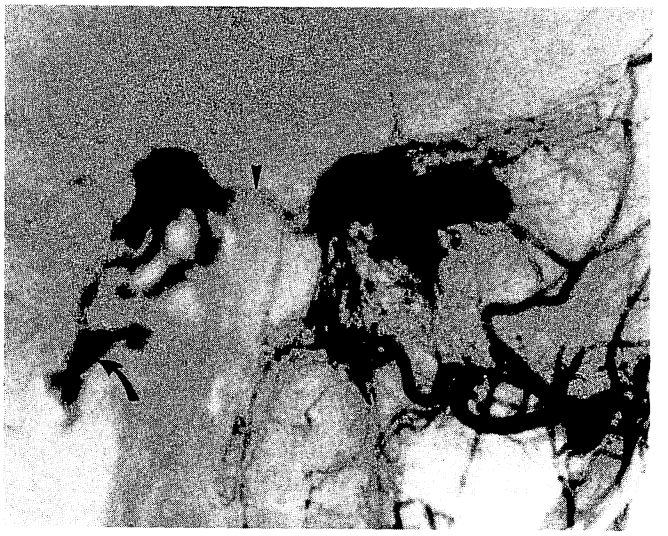
be the initial symptoms of CRVO. As in our case, it should be noted that the CRVO may occur even after temporary clinical improvement.

In a dural CCF, the elevated venous pressure in the cavernous sinus and tributaries can cause retinal ischemia due to the decreased retinal perfusion pressure, even without occlusion of the central retinal artery [16]. The degree of retinal ischemia depends upon whether the CRVO is of the venous stasis or hemorrhagic type [20]. The raised intraocular pressure may contribute to hypoperfusion of the retina, resulting in hemorrhagic retinopathy. For this reason, it is necessary to control the intraocular pressure to prevent this complication [12].

"Carotid–jugular compression" is the accepted treatment for a dural CCF, except in cases with cortical venous drainage and progressive visual decline. This maneuver may facilitate thrombosis formation in the cavernous sinus. Halbach et al [3] showed that there were no complications in 23 patients treated with carotid–jugular compression, and seven patients experienced complete cure. However, Miki et al [8] reported three patients with dural CCF who experienced retinal hemorrhaging during treatment by the Matas procedure. The retinal hemorrhaging in their cases seemed to be due to CRVO. It should be noted that carotid–jugular compression can produce thrombosis formation in the cavernous sinus and in the CRVO due to the intentionally reduced retinal perfusion pressure.

Embolization for treatment of a dural CCF is now possible thanks to improvements in microcatheters, guidewires, and embolic agent technology [2,19]. With the increased popularity of this treatment modality, possible complication by CRVO should be recognized. Even transarterial embolization may induce thrombosis in the cavernous sinus, which can cause CRVO, as shown in our case. Transvenous embolization means artifical occlusion of the cavernous sinus, ie, the outflow of the central retinal vein, and increases the risk of this complication [4]. In order to reduce the likelihood of CRVO, the embolic materials should be placed only within the site of the fistula in the cavernous sinus.

The transvenous approach is usually through the inferior petrosal sinus or the superior ophthalmic vein [2,17]. A transvenous approach through the superior ophthalmic vein is indicated when drainage from the cavernous sinus is solely to the superior ophthalmic vein and catheterization of the cavernous sinus through the inferior petrosal sinus is impossible. We think that "cut down" of the superior ophthalmic vein for the introduction of a catheter should be avoided, because sacrificing this vein increases the possibility of deterioration of the ocular symptoms when embolization is incomplete or unsuccessful. Instead, a microcatheter can be introduced into the cavernous sinus through



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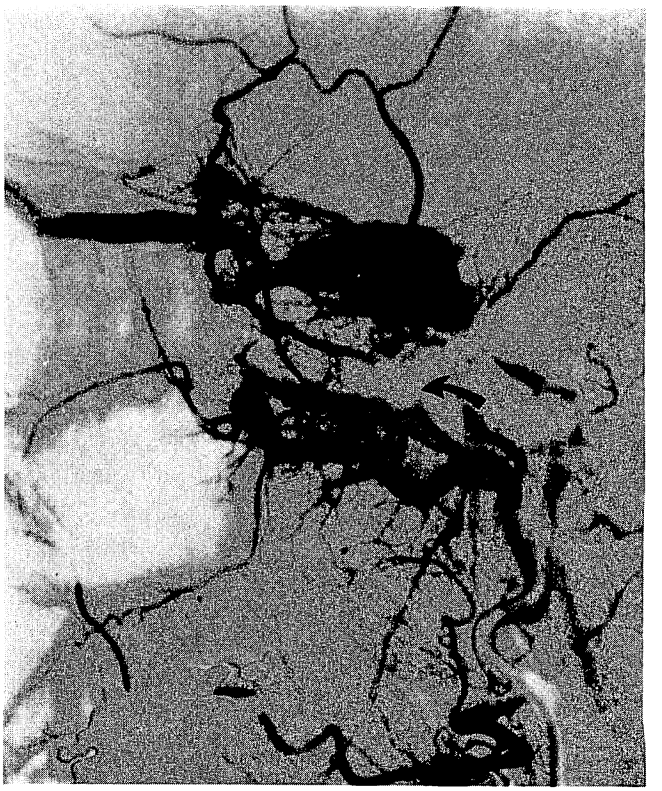

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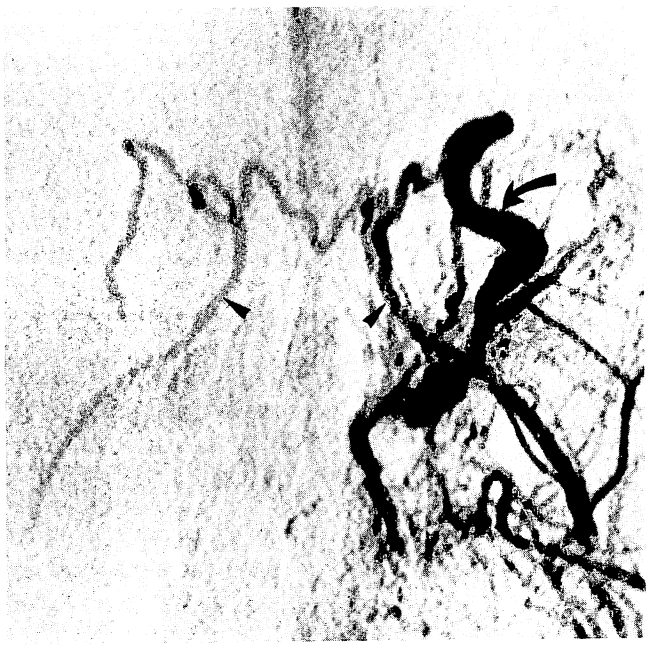
Figure 1. *Left external carotid injection carried out in June 1989.* **(A)** *Anteroposterior view. The dural carotid–cavernous sinus fistula is clearly depicted. The drainage routes are mainly to the left superior ophthalmic vein and to the right inferior petrosal sinus (arrow) through the intercavernous sinus (arrowhead).* **(B)** *Lateral view. The left middle meningeal artery is contributing to the dural fistula (arrow).*



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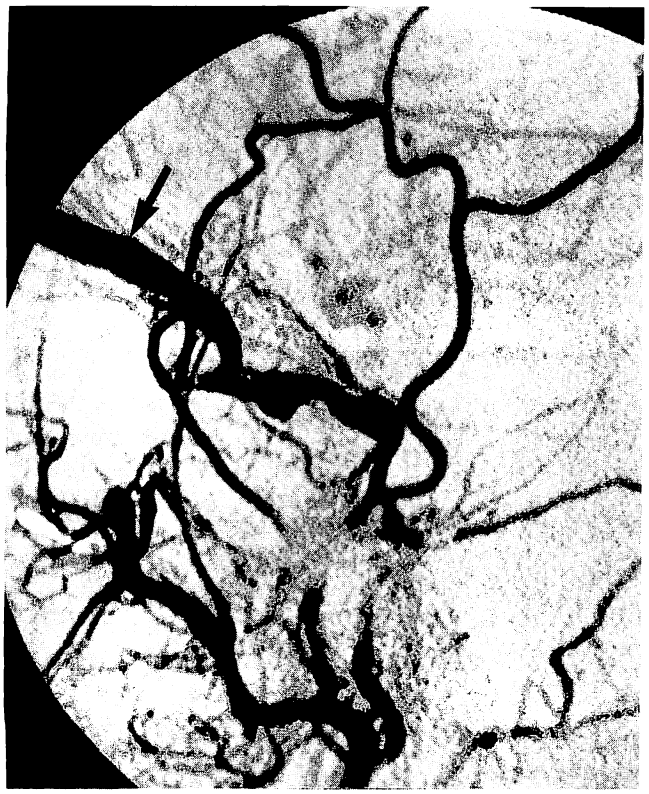

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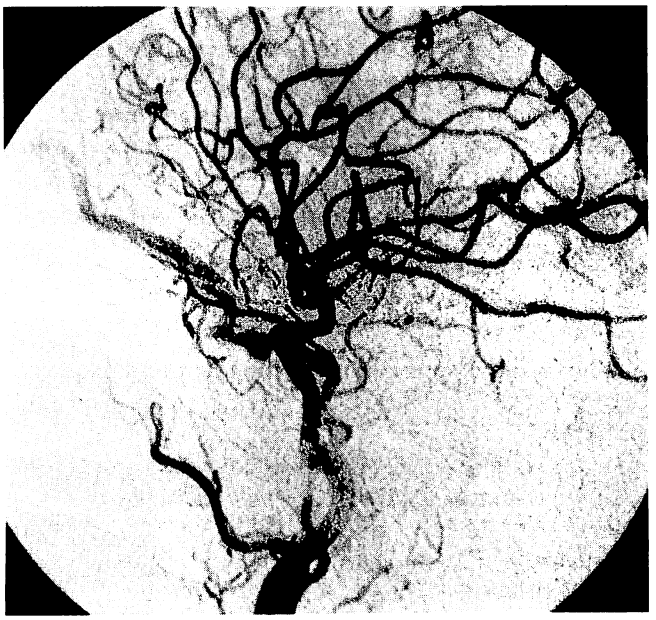

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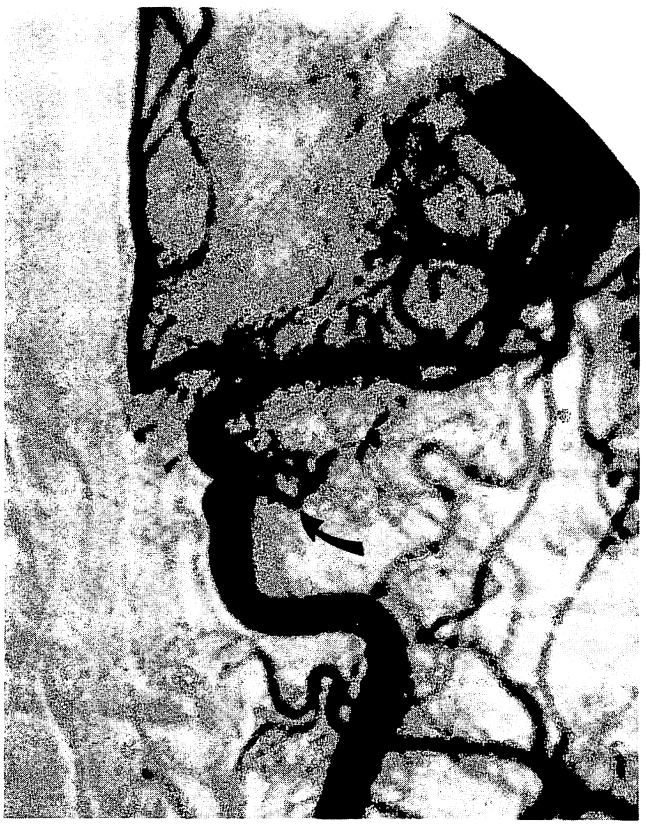
Figure 2. *Particulate embolization was carried out in early July 1989.* **(A, B)** *Anteroposterior and lateral views of the left external carotid injection before embolization. The drainage route had changed solely to the left superior ophthalmic vein (arrows) and then to the bilateral angular veins of the face (arrowheads).* **(C)** *Lateral view of the postembolization left common carotid injection. Embolization was carried out of the left middle meningeal artery and the left ascending pharyngeal artery. Shunted blood in the left superior ophthalmic vein is markedly reduced and the meningohypophyseal artery of the left internal carotid artery is contributing to the dural fistula.*

the superior ophthalmic vein without "cut down" of this vein [7].

As shown in Table 1, CRVO occurred after endovascular treatment in five cases, including our case [4,20]. The CRVO occurred in the course of the natural history of a dural CCF with conservative therapy in nine cases [1,10–12,18], and transarterial embolization improved the ocular symptom in one case [16]. A review of the literature showed that CRVO associated with dural CCFs may be more common than previously thought. In the course of the many possible therapeutic modalities, including conservative therapy, all patients should be instructed about the possibility of CRVO, observe for changes in visual acuity, and undergo frequent ophthalmologic examinations [2,12]. Early detection of CRVO would allow early intervention, which might prevent deterioration of vision. Anticoagulation therapy and fibrinolytic therapy might be effective in the very early stages of CRVO. Control of the intraocular pressure is also indispensable. More aggressive therapy such as embolization might also be effective in some cases. In conclusion, dural CCFs should be treated and followed taking into account possible complication by CRVO.



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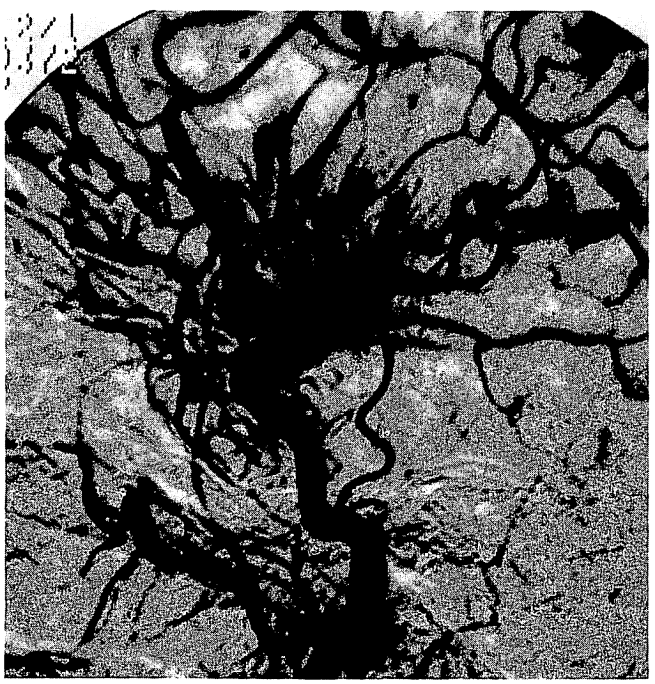

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Figure 3. *Left common carotid injection carried out in December 1989, when the patient developed the central retinal vein occlusion.* **(A, B)** *Anteroposterior and lateral views show faint opacification of shunted blood in the left superior ophthalmic vein (arrows). The left ophthalmic artery is normal. The left middle meningeal artery embolized in July has recanalized (arrowhead).*

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