Successful surgical treatment of bilateral carotid-cavernous fistulas

Case report

FRANCES K. CONLEY, M.D., RICHARD D. HAMILTON, M.D., AND YOSHIOS HOSEBUCHI, M.D.

Department of Surgery, Division of Neurosurgery, Stanford University School of Medicine, and Department of Neurological Surgery, University of California School of Medicine, San Francisco, California

The authors report a case of traumatic bilateral carotid-cavernous sinus fistulas, successfully treated by surgical electrothrombosis of both cavernous sinuses.

KEYWORDS: carotid-cavernous fistula, surgical electrothrombosis, angiography

The occurrence and recognition during life of bilateral carotid-cavernous sinus fistulas is very rare. In 1971, Voigt, et al., found four cases of angiographically verified bilateral lesions in the literature, and added one further case. To our knowledge, only three subsequent cases have been reported. Of these eight cases three were of spontaneous onset, four of traumatic origin, and one developed after very minimal trauma in a patient with bilateral internal carotid artery aneurysms. Only two were treated successfully by surgery.

Case Report

This 14-year-old girl was admitted, comatose and in severe respiratory distress, following an automobile accident in September, 1972. Blood was coming from both nostrils and both external auditory canals. The right pupil was dilated and fixed to light; the left pupil reacted sluggishly and the eye deviated inward with occasional nystagmoid movements.

Examination. No papilledema and no cranial bruits could be found. Deep painful stimulation produced nonpurposeful movements of the left arm and leg only. Plain skull films showed multiple linear fractures in the left temporoparietal area. A left carotid arteriogram was performed when the patient's respiratory status had stabilized; it showed no midline shift, but revealed disruption of the cavernous portion of the internal carotid artery (ICA) with opacification of the cavernous sinus and early filling of the left internal jugular vein. The right side was not studied.

Over the next 6 days the patient required tracheostomy, and developed bilateral orbital chemosis and a right orbital bruit. Nine days...
after injury bruits were present over both eyes, and on the twelfth day, cerebral arteriography by femoral catheter was performed. The left-sided injection showed immediate filling from the intracavernous portion of the ICA of large, tortuous venous structures in the region of the left cavernous sinus. In addition, there was cross filling into the right anterior and middle cerebral arteries and demonstration of a right carotid-cavernous fistula (Fig. 1 left). This was confirmed by a right ICA injection; the right injection did not fill the left-sided fistula (Fig. 1 right). A vertebral study failed to demonstrate either fistula, although flash filling of both right and left anterior circulations was noticed.

First Operation. One month after injury the patient was alert, moving all extremities, and asking questions. Bilateral orbital chemosis and proptosis persisted as did the loud bruit. Seven weeks after the accident a right frontal craniotomy was performed with intracranial ligation of the right ICA above the origin of the ophthalmic artery. The right cervical carotid artery was exposed and the ICA ligated 1.0 cm above the bifurcation, after unsuccessful attempts were made to pass muscle emboli into the fistula. Following surgery there was some improvement of symptoms. Arteriography showed no filling of the right fistula from the left ICA or the left vertebral artery, but showed persistence of the right-sided defect by feeders from the right external carotid artery through communication with the ophthalmic artery as well as by direct meningeal branches.

The patient was transferred to the University of California Medical Center in November, 1972, for direct surgical obliteration of the bilateral fistulas. At the time of transfer, she was blind in the right eye, had a visual acuity of 20/400 in the left eye, and severely limited extraocular motion.
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Fig. 2. Left: Left carotid arteriogram showing the complete occlusion of the left carotid cavernous fistula. The arrow indicates the persistent false aneurysm of the cavernous carotid artery. Right: Right external carotid arteriogram, lateral projection, after the entrapment procedure and the electrothrombosis of the left carotid cavernous fistula. The fistula is primarily fed through the right ophthalmic artery. The single arrow indicates the clip placed on the intracranial carotid artery. The double arrows represent the copper needles in the left cavernous sinus.

bilaterally. Loud bruits and severe bilateral chemosis persisted, as did a mild right hemiparesis. Fifth nerve function was decreased over the right first and second divisions.

Second Operation. A left frontal craniotomy was done, the left cavernous sinus exposed under direct vision, and the location of the fistula in the posterior portion of Parkinson's triangle documented by intraoperative angiography. Six feet of 2/1000-inch diameter copper wire was inserted directly into this area of the cavernous sinus through thin-walled hypodermic tubing; this was followed by several short 26 to 29-gauge copper needles. Direct anodal current (0.02 to 0.5 mA) was applied to the ends of the wire and needles for 60 to 120 seconds each to initiate thrombosis. Following the insertion of each needle or wire, the progression of the thrombosis was monitored with intraoperative angiograms. Diminished but persistent posterior filling of the fistula was stopped by slowly injecting a 50:50 mixture of liquefied bone wax and mineral oil into the midportion of the cavernous sinus inferior to the cavernous carotid artery. Postoperative angiography showed closure of the left fistula (Fig. 2 left), but persistence of the right, which was fed by retrograde flow from the right external system (Fig. 2 right).

Third Operation. Three weeks later the right-sided craniotomy was reopened, and that fistula was successfully obliterated by the same direct cavernous sinus coagulation technique (Fig. 3).

Postoperative Course. Seven months after injury, in April, 1973, a follow-up angiogram indicated complete occlusion of both carotid-cavernous fistulas. The patient remains blind in her right eye and the eye is deviated medially with limited abduction and upward gaze. Vision in the left eye has returned to 20/50, uncorrected, with full extraocular movements. Hypalgesia, present before surgery, persists in the first division on the right. The patient is alert, active, and pursues the normal existence of a healthy teenager.

Discussion

The diagnosis during life of bilateral carotid-cavernous fistulas can only be made by angiography as it is well known that a single fistula can produce bilateral symptomaticity. However, the presence of
Fig. 3. Right external carotid arteriogram shows the complete occlusion of the right carotid cavernous fistula following the electrothrombosis procedure. The arrows indicate the copper needles in the cavernous sinuses. The wires are totally subtracted in this x-ray preparation.

### TABLE 1

Summary of nine cases of angiographically demonstrated bilateral carotid-cavernous fistulas

<table>
<thead>
<tr>
<th>Author, Year</th>
<th>Etiology</th>
<th>Treatment</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mason, et al., 1954</td>
<td>trauma</td>
<td>surgery</td>
<td>residual bilateral abducens palsy; hypalgesia divisions 1, 2 of CN V</td>
</tr>
<tr>
<td>Zander, 1959</td>
<td>trauma</td>
<td>surgery</td>
<td>residual bilateral abducens palsy</td>
</tr>
<tr>
<td>Hellner, 1962</td>
<td>minor trauma; bilateral ICA</td>
<td>none mentioned</td>
<td>unknown</td>
</tr>
<tr>
<td>Voigt, et al., 1971</td>
<td>spontaneous</td>
<td>spontaneous occlusion after angiography</td>
<td>patient well; normal arteriogram</td>
</tr>
<tr>
<td>Friedmann, et al., 1970</td>
<td>trauma</td>
<td>none</td>
<td>arteriogram 13 yrs after accident; bilateral exophthalmus, decreased vision</td>
</tr>
<tr>
<td>Stolpmann, 1972</td>
<td>spontaneous</td>
<td>intermittent carotid compression</td>
<td>patient well; rt arteriogram showed no fistula; It arteriogram not done</td>
</tr>
<tr>
<td>Stolpmann, 1972</td>
<td>spontaneous</td>
<td>intermittent carotid compression</td>
<td>no long-term follow-up; slightly improved at 3 mos died</td>
</tr>
<tr>
<td>Kubin &amp; Ortner, 1973</td>
<td>trauma</td>
<td>none</td>
<td>blind rt eye; hypalgesia division 1, CN V, rt</td>
</tr>
<tr>
<td>Conley, et al., 1975</td>
<td>trauma</td>
<td>surgery</td>
<td></td>
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bilateral symptoms and/or the persistence of a bruit after surgical therapy should alert one to the possibility of bilateral fistulas. In two of the eight cases previously reported, the second fistula was recognized only when treatment of the first side failed, and in another only during routine evaluation of the intracranial circulation prior to surgery. The arteriographic diagnosis is made early in the arterial phase when contrast is seen filling the cavernous sinus directly from the siphonous portion of the internal carotid artery. Also, those veins draining into the cavernous sinus may show a reversal of flow.

The goal of any therapeutic measure is to effect closure of the arteriovenous shunts. Stolpmann reported the alleviation of symptoms in two cases of spontaneous bilateral carotid-cavernous fistulas by daily intermittent alternating carotid compression, carried out over several months. The case of spontaneous onset reported by Voigt, et al., occluded spontaneously following cerebral arteriography; repeat angiography 1 year later failed to demonstrate either fistula.

Two of the eight previously reported cases were treated surgically, and both survived (Table 1). The patient of Mason, et al., underwent a right-sided “trapping” procedure, followed by ligation of the left internal and common carotid arteries; the patient has a residual bilateral abducens palsy and left first and second fifth nerve sensory deficit. Zander's patient had bilateral muscle embolization of the fistulas when it became apparent during an attempted right-sided trapping procedure that bilateral fistulas were present. The only deficit remaining was bilateral abducens palsy.

The risks of mortality and morbidity with bilateral carotid occlusion and/or embolization are considerable. Obliteration of the involved venous structures with closure of the fistulas and preservation of the carotid arterial circulation offers an attractive alternative. The technique has been described previously; the copper wire used is soft and small in diameter and will not damage the neurovascular structures in the cavernous sinus. The larger diameter copper needles, however, could skewer the cranial nerves and lead to permanent damage. Therefore, by using a large quantity of the copper wire, and facilitating the occlusion with the bone wax-mineral oil mixture, the use of a large quantity of copper needles is minimized.

The successful outcome of this case indicates the value of induced cavernous sinus thrombosis as the definitive procedure for treatment of single or bilateral carotid-cavernous fistulas.

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References


Address reprint requests to: Frances K. Conley, M.D., Stanford University School of Medicine, 300 Pasteur Drive, Stanford, California 94305.