Dural Malformations with Ophthalmic Manifestations: Results of Particulate Embolization in Seven Patients

Seven patients with proptosis, chemosis, diplopia, and elevated intraocular pressure were found to have dural arteriovenous malformations with drainage into the cavernous sinus and superior ophthalmic vein. These patients had sufficient symptoms to justify therapeutic embolization with polyvinyl alcohol. Five of seven patients had complete resolution of their ophthalmic symptoms after embolization; the other two had transient improvement. Follow-up was 9–29 months. When 50% or more of the vessels supplying a malformation were embolized, complete relief of symptoms was obtained. Particulate embolization is an effective therapeutic approach in patients with significant ophthalmologic symptoms from dural malformations.

Dural malformations are well described abnormal communications between dural arteries and venous sinuses [1–5]. Those patients in whom the malformation is between dural blood vessels and the cavernous sinus have symptoms referable to the eye and orbit. In most patients these clinical abnormalities are mild and, left untreated, may undergo spontaneous resolution [6]. However, we have observed another group of patients who required radiologic intervention because of significant visual loss, diplopia, and proptosis. We describe the ophthalmologic manifestations, radiologic anatomy, and results of treatment by embolization of cavernous sinus dural malformations in seven patients.

Materials and Methods

Our series comprised seven patients, five women and two men, aged 40–73 years. Each patient’s symptoms were of at least 3 months duration. The most common orbital findings were conjunctival injection (seven patients), proptosis (five), pain (six), diplopia (four), and asymmetrically increased intraocular pressure (six). Three patients had an isolated cranial nerve palsy, and this involved the fourth, sixth, and seventh cranial nerves. Other clinical signs included visual field defects (three of seven), decreased visual acuity (three), ptosis (three), and bruit (three). Symptoms were localized predominantly to one eye (right eye, six patients; left eye, one).

All patients were embolized using polyvinyl alcohol (PVA) (Unipoint, High Point, NC) [7, 8]. The PVA was prepared by suspending the prepackaged particles (149–250 μm) in saline and then mixing the suspension in a blender for about 15 min. The PVA suspension was poured into 50 ml vials, sterilized, and stored. This technique enables rapidity of suspension with contrast material. When needed the saline is decanted and replaced by iothalamate meglumine (Conray 60, Mallinckrodt) with 2 ml of 25% albumin added to help keep the PVA in suspension. A 7 French Cook Check-Flo Sheath (Cook, Bloomington, IN) was placed in the femoral artery to facilitate rapid atraumatic catheter exchange. The sheath was attached to a constant perfusion of heparinized saline (5000 U/500 ml). The USC 7 French Berenstein catheter (UCSI, Bilerica, MA) or a 4 or 5 French Hanafee “carotid-shaped” catheter (B-D, Rutherford, NJ) was used in conjunction with a 0.32, 0.35, or 0.38 inch (0.81, 0.89, or 0.97 cm) Bentzon wire (Cook) for selective catheterization [9]. Introduction of the catheter by direct carotid puncture was useful when aortic arch tortuosity (two patients) precluded selective transfemoral catheter placement. Our approach in this situation consists of imaging the carotid
bifurcation at the time of the diagnostic study to search for plaques, stenosis, and the level of the carotid bifurcation. The direct carotid puncture was made using an 18 gauge Potts-Cournand needle. A 4 French Hanafee catheter, steamed to a shape similar to an H-1, was then exchanged over a 0.32 inch (0.81 cm) Bentson wire. The flexible wire and soft catheters caused little vascular spasm and enabled selective catheterization.

The introduction of PVA was increased considerably by placing an 18 gauge Potts-Cournand needle directly into the proximal catheter end. This needle in conjunction with syringe agitation permitted large amounts of PVA to be delivered into the vessel without clumping of PVA in the hub of the syringe. We have since developed a PVA introducer (Cook) that accomplishes the same purpose (fig. 1).

Our embolization protocol consists of performing a diagnostic examination using cut film before embolization. The embolization itself is monitored and recorded on a Diasonics digital imager (512 matrix). This provides on-line digital subtraction and obviates, in most cases, hand-subtracted film. Regardless of which side is symptomatic, angiography usually includes both internal and external carotid arteries. In addition, selective catheterization of the appropriate vessels is also performed. If we were unable to selectively catheterize the external vessels during the diagnostic study because of aortic tortuosity, then common carotid arteriograms were obtained (two patients). In these cases selective external catheterization and angiography were accomplished at the time of the therapeutic study after direct carotid puncture.

These procedures can be painful. Our analgesic approach initially was to use droperidol 2.5 mg intravenously and supplement it with morphine 2.5 mg intravenously. We now have an anesthesiologist in attendance during embolizations and have been using Pentothal drips (500 mg in 250 ml D5/W at 1–2 ml/min). This has markedly increased patient tolerance of the procedure.

**Results**

Our follow-up period was 9–29 months. In five of seven patients embolization produced complete relief of symptoms. In the other two, only partial relief was obtained and the malformation remained patent. In three patients incomplete embolization did not preclude closure of the malformation. In one patient who had embolization of the unilateral external feeders, another angiogram 2 months later demonstrated disappearance of the supply from the opposite, nonembolized meningeohypophyseal branches (fig. 2). In the other two patients fistula closure was inferred from complete relief of symptoms.

Case 1 represents a most unusual situation. The patient was symptomatic for 3 months but then experienced an acute exacerbation over a 2 day period. Angiography revealed that the malformation was supplied solely from the accessory meningeal artery and that the anterior aspect of the superior ophthalmic vein was occluded. This thrombosis caused the blood to be shunted predominantly through venous collaterals around the orbit (fig. 3).

The three patients who displayed a cranial nerve palsy at presentation experienced resolution after embolization. Seventh nerve palsy is an exceedingly rare manifestation of carotid-cavernous fistulas, having been described only on two previous occasions [10, 11]. Increased blood flow through the cavernous sinus into the inferior petrosal sinus producing retrograde engorgement of the vein from the internal auditory

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**Fig. 1.**—PVA introducer. This 18 gauge introducer is held directly on catheter hub and permits infusion of large quantities of PVA. Short segment of introducer projects into syringe and significantly decreases clumping of PVA.

**Fig. 2.**—Case 3. A, Right common carotid injection. blush from meningeohypophyseal trunk (arrow). B, 2 months after contralateral embolization. Little, if any, vascular malformation.
meatus has been postulated to produce the seventh nerve dysfunction [11]. In general, all symptoms had resolved completely 12 weeks after embolization, although improvements in the appearance of the eye were manifest 24 hr after embolization.

In two patients (cases 5 and 6) the initial embolizations were not complete, and both of these patients refused a second attempt at fistula closure. In two other patients (cases 3 and 4) incomplete embolization secondary to poor tolerance of the procedure necessitated repeat embolization. In these four situations (cases 3–6), many vessels supplied the malformations, and the duration of the embolization was directly related to the number of vessels supplying the malformation. Patient tolerance decreased as the embolization became prolonged, and the patients were given the option of a second attempt at another time. One patient experienced transient hemiparesis (<24 hr). This was most likely the result of the dislodgment of a small plaque from the bifurcation during catheter repositioning. The most common postembolization complaint was pain and difficulty in opening the mouth fully. These symptoms usually resolved over 1 week.

In six of seven cases multiple vessels supplied the malformation (fig. 4). The vessels contributing to the malformation included the middle meningeal artery, the accessory meningeal artery, the artery of the foramen rotundum, the ascending pharyngeal artery, the occipital artery, and branches of the meningohypophyseal trunk. Despite the fact that symptoms were unilateral, bilateral vascular supply was demonstrated in six of seven cases. Venous drainage occurred predominantly retrograde via the superior ophthalmic vein. Other routes of venous outflow included the inferior petrosal sinus, pterygoid plexus via the foramen ovale, and superficial cortical veins.

Two patients (cases 1 and 4) in whom all vessels that supplied the malformation were embolized had resolution of their symptoms. Three other patients (cases 2, 3, and 7) who had incomplete embolization also experienced symptom relief. The arteries not embolized in these cases included the meningohypophyseal branches (cases 2, 3, and 7) as well as supply from the contralateral side (cases 2 and 7). In those
Discussio

meningeal branches of the internal and/or external carotid artery are often insignificant and require no therapy [2, 12]. Spontaneous thrombosis, particularly after angiography, is a well described entity [13-15]. The patients in our series had significant ophthalmologic problems that caused them to seek medical attention and to accept the risk of diagnostic angiography and embolization. These findings included orbital pain, diplopia, visual loss, proptosis, bruit, and asymmetrically elevated intraocular pressure. Furthermore, the patients' symptoms had persisted for at least 3 months. When, or if, a spontaneous carotid-cavernous fistula will close is problematic. For this reason, when progressive symptoms necessitate therapeutic intervention, particulate embolization is a reasonable alternative to either conservative therapy or surgery [16, 17].

Although six of seven patients had bilateral vascular supply, only one orbit was significantly involved. This may be related to the pattern of venous drainage from the cavernous sinus and areas of venous thrombosis. Obstruction of the superior ophthalmic vein in its distal segment produced a fulminant situation (fig. 3). The patient's condition deteriorated over 2 days with pronounced elevation of intraocular pressure to 37 mm Hg as well as visual loss, pain, and 11 mm of proptosis. The angiogram revealed marked orbital venous collaterals representing the route of decompression of the malformation. This clinical situation requires immediate treatment. With the exception of this case we could discern no correlation between symptomatology and collateral venous drainage. When retrograde venous drainage occurred through the superior ophthalmic vein orbital symptomatology was present and alternative routes of venous drainage did not necessarily infer decreased orbital findings.

Our approach to the therapy of dural cavernous malformations involves selective catheterization of the appropriate vessels, thereby unambiguously displaying whether or not a particular artery supplies the malformation. In addition, potential hazardous anastomoses are demonstrated (fig. 5) [18]. Selective catheter placement affords less opportunity for embolic reflux and occlusion of noncontributory vessels. To deliver large amounts of PVA a Potts-Cournard needle or our modification of it as an introducer is most effective. This technique decreased the clumping of PVA in the needle hub and significantly increased the speed of the procedure. When aortic tortuosity precludes selective catheterization from the femoral route a direct carotid puncture and catheterization using a 4 French Hananee catheter appears to be safe and effective.

When our results are analyzed with respect to the arteries that supply the malformation compared with the number of vessels that were embolized, it becomes apparent that successful treatment of the lesion does not necessarily require embolization of all the feeders. This was also appreciated by Vinuela et al. [19], who reported complete clinical cure in five patients 6 months after incomplete embolization. In three of five patients with complete relief of symptoms, embolization was not complete. In these cases the contralateral feeding vessels were not embolized initially, and during clinical follow-up it became apparent that a repeat procedure was unnecessary. In our case 3, a repeat angiogram demonstrated that a meningo-hypophyseal anastomosis that had previously supplied the malformation was no longer seen. Thus, if greater than 50% of the arteries supplying the fistula were embolized, there was an excellent chance of complete closure. Further-

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Fig. 4.—Arteries contributing to dural fistula are on left and are divided into left (L) and right (R)-sided supply. Gray columns indicate whether or not a particular vessel supplied malformation. White columns represent those arteries embolized and not embolized. Total number of arteries treated per case is also stated. Number of embolization attempts per case is at bottom.

Fig. 5.—Case 4. Anastomosis between ascending pharyngeal and vertebral arteries (arrow). Filling of fistula. Selective catheterization permits recognition of important anastomosis.

Discussion

Dural malformations involving the cavernous sinus and meningeal branches of the internal and/or external carotid artery are often insignificant and require no therapy [2, 12]. Spontaneous thrombosis, particularly after angiography, is a well described entity [13-15]. The patients in our series had significant ophthalmologic problems that caused them to seek medical attention and to accept the risk of diagnostic angiography and embolization. These findings included orbital pain, diplopia, visual loss, proptosis, bruit, and asymmetrically elevated intraocular pressure. Furthermore, the patients' symptoms had persisted for at least 3 months. When, or if, a spontaneous carotid-cavernous fistula will close is problematic. For this reason, when progressive symptoms necessitate therapeutic intervention, particulate embolization is a reasonable alternative to either conservative therapy or surgery [16, 17].
more, embolization of only the symptomatic side could have resulted in complete closure (cases 2, 3, and 7). This suggests that the particulate material serves as a nidus for perpetuation of thrombosis throughout the cavernous sinus or that the abnormal hemodynamics of the fistula are sufficiently altered so that remaining abnormal venous drainage is no longer clinically significant. Instilling enough particulate material as well as decreasing flow through the fistula helped promote its closure.

Particulate embolization is a safe and effective method of treating dural fistulas of the cavernous sinus in patients with potentially disabling ophthalmic symptoms. Embolization appears to be a reasonable therapeutic choice when dural vascular malformations produce fulminant symptoms and threaten vision.

ACKNOWLEDGMENT

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REFERENCES

1. Newton TH, Greitz T. Arteriovenous communication between the occipital artery and transverse sinus. Radiology 1966;87:824-828