Paraplegia due to posttraumatic pelvic arteriovenous fistula treated by surgery and embolization

Case report

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A case is presented in which a posttraumatic pelvic arteriovenous fistula caused progressive paraplegia because of voluminous shunting into the epidural venous system. Surgical ligation and transcatheter embolization of major and minor arterial feeders decreased shunt flow sufficiently to permit direct embolization of the fistula by an injectable plastic. This combined approach may allow obliteration of unresectable acquired or congenital arteriovenous malformations.

KEY WORDS arteriovenous fistula traumatic arteriovenous fistula embolization paraplegia

ARTERIOVENOUS malformations (AVM's) of the spinal cord produce neurological symptoms by diverting or "stealing" blood from the spinal cord, by compressing the cord with dilated arterial and venous channels, or by producing chronic venous hypertension in the cord.1,8,12 Neurological dysfunction resulting from extra-spinal arteriovenous fistulas is rare.6

In this case report of progressive paraplegia resulting from a longstanding acquired extra-spinal arteriovenous fistula, we describe a combined surgical-embolic approach to a difficult management problem. Surgical ligation of major feeders was followed by transcatheter embolization of the remaining blood supply. Finally, a rapidly polymerizing plastic was injected directly into the fistula. This sequence completely obliterated the fistula with alleviation of symptoms and partial restoration of neurological deficits.

Case Report

This 27-year-old Mexican American man was admitted to the National Institutes of Health (NIH) Clinical Center for evaluation of progressive bilateral spastic paraparesis. In 1960, he sustained a gunshot wound to the right buttock with exit in the midline over the symphysis pubis. A loud bruit subsequently developed in the right lower abdomen and arteriography demonstrated an arteriovenous fistula involving the right internal iliac vessels. From 1961 to 1964, four separate attempts were made to control this lesion by ligation of feeding vessels. The fistula recurred following each procedure, and subsequent arteriograms showed progressive enlargement of the arteriovenous aneurysm.

In 1971, the patient developed fatigue, weakness, and decreased sensation in the lower extremities. By mid-1974, the patient required crutches for walking; he had difficulty in achieving an erection, and had lost normal bowel and bladder function. A myelogram revealed multiple, serpentine, filling defects in the lumbar subarachnoid space, and displacement of the caudal sac to the left by an epidural mass (Fig. 1). By 1976, progressive lower extremity weakness prevented his ambulation even with crutches. He was referred to NIH at this time.

Examination. Physical examination revealed a loud bruit in the right lower quadrant of the abdomen and over the sacrum. There was decreased sensation bilaterally to pinprick at the L-4 vertebral level. Ankle jerks were absent and a Babinski sign was present on
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FIG. 2. Selective inferior mesenteric arteriogram reveals a massive collection of dilated vascular channels in the right side of the pelvis (small arrows) draining mainly into enlarged epidural veins (large arrows) which are responsible for displacement of the caudal sac.

FIG. 1. Serpentine filling defects (arrowheads) due to enlarged vessels within the subarachnoid space. The caudal sac is deviated to the left (arrows) by an extradural mass.

the left. Bilateral lower extremity weakness was present, greater on the left, and the gait was spastic and wide-based. Anal sphincter tone was decreased, and a large pulsating vessel was felt along the right lateral wall of the rectum. Pulses in the lower extremities were equal and full. No edema or varicosities were present. No cardiovascular deficits at rest were demonstrable.

Angiography showed a huge right pelvic arteriovenous fistula fed by the inferior mesenteric artery (Fig. 2), and many collateral transpelvic branches through the bed of the prostate and base of the bladder (Fig. 3). The main venous drainage was through massively enlarged epidural veins (Figs. 2 and 3), presumably because the right internal iliac vein had been ligated at a previous exploration.

Operations. The abdomen was explored through a lower midline incision. An aneurysm was identified deep in the right pelvis with its posterior wall adherent to the upper sacral nerve roots. The right ureter coursed over the superior wall of the malformation and was densely adherent to it. The inferior mesenteric and left internal iliac, obturator, and pudendal arteries supplied the arteriovenous fistula, and were grossly enlarged. These vessels were serially isolated, ligated, and divided, but the aneurysm could not be resected because of the intimate association with sacral nerve roots. To assure control of all feeding vessels, transection of dense fibrous tissue containing the terminal right ureter was necessary. This required a transureterostomy. The medial wall of the aneurysm was closely bound to the bladder which was displaced by scarring from previous procedures. Total blood replacement was 65 units.
In the postoperative period, the patient developed signs and symptoms of continued blood loss and was re-explored. Multiple small bleeding points along the site of the dissection were isolated and controlled. After the second operation there was marked weakness of dorsiflexion of the right foot with a spotty loss of sensation in the sacral distribution of the right side. This was presumed due to sacral nerve injury sustained in the course of controlling the bleeding vessels. Renal function remained normal throughout the postoperative period.

Postoperative Course. Five weeks later an arteriogram revealed three small persistent arterial feeders from the left internal iliac and right circumflex iliac arteries (Fig. 4). Each was embolized with Gelfoam (Fig. 5). Flow through the fistula was greatly reduced, as demonstrated by its prolonged opacification at arteriography. The fistula was then directly punctured with a No. 23 Chiba needle and 1.5 ml of a rapidly polymerizing plastic (isobutyl cyanoacrylate) was injected directly into it (Fig. 6). The patient had no complications from this procedure and was discharged 2 days later.

Two months postoperatively, motor function and sensation had gradually returned to the left leg, with the exception of a persistent Babinski response. On the
right side, a foot drop with continued spotty sensory loss in the sacral distribution persisted. A cystometrogram revealed a mildly flaccid, neurogenic bladder. A second postoperative arteriogram revealed a few residual feeders. These were embolized, followed by a second direct injection of 1.25 ml of isobutyl cyanoacrylate into the venous aneurysm in the right pelvis. Six months postoperatively two small additional feeders from presacral vessels were embolized and the aneurysmal portion of the malformation was occluded.

Fig. 4. Postoperative aortogram. *Left:* Arterial phase shows dramatic diminution in size, but persistent opacification of the fistula (*small arrows*) by two left presacral and one right circumflex femoral feeders (*large arrows*). *Right:* Venous drainage into the epidural plexus still predominates.

Fig. 5. Selective arteriograms of presacral feeder. *Left:* Before embolization there is persistent opacification of right pelvic fistula (*arrows*). *Right:* After Gelfoam embolization, proximal obstruction of this feeder is demonstrated (*arrow*) with reflux into the superior gluteal artery.
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obliterated a third time with 0.5 ml isobutyl cyanoacrylate.

The patient’s neurological function had not improved over the status observed 2 months after the operation. Fourteen months postoperatively angiography failed to demonstrate any evidence of the fistula. Two years after the operation the left-sided Babinski sign and the right-sided foot drop and spotty sensory loss in the sacral distribution remained. However, in marked contrast to the severe lower-extremity weakness which prevented his walking before the operation, the patient was able to walk with the aid of a cane and a right-foot brace.

Discussion

The pathogenesis of progressive cord dysfunction in congenital AVM’s of the spinal cord is not fully established. Cord compression by dilated vessels and cord ischemia secondary to “steal” by the arteriovenous fistula have both been proposed. Aminoff, et al., have suggested, on the basis of analysis of a large number of patients, that venous hypertension may be the principal mechanism, a theory fitting nicely with the angiographic findings in our patient. Extra-spinal arteriovenous fistulas may also rarely be responsible for progressive spinal cord dysfunction.

In the patient reported here, the failure to demonstrate an enlarged anterior spinal artery and the diversion of significant venous drainage into the epidural plexus after surgical obliteration of the usual pelvic venous pathways suggest that venous hypertension and/or compression by enlarged epidural veins were responsible for this patient's progressive paraplegia. Regardless of the pathophysiological mechanism, control of the progressive myelopathy in our patient required obliteration of the pelvic arteriovenous fistula. The standard treatment for acquired arteriovenous fistulas involving major vessels is early exploration and immediate repair. The gratifying results achieved with this approach in the Korean and Vietnamese conflicts are contrasted to the earlier rather disappointing experiences using ligation and excision as the primary treatment.

When arteriovenous fistulas are treated by simple ligation of the feeding arteries without repair or resection of the fistulous communication, new arterial feeders invariably develop. The blood supply to the fistula often becomes devous and inaccessible to direct catheterization. Our patient illustrates this problem of an inappropriate initial surgical approach. Because the right iliac arteries originally supplying the fistula had been ligated, access to the fistula with a catheter for oblitative treatment by means of a rapidly polymerizing plastic was technically impossible. Our approach was as follows: 1) to obliterate the major arterial feeders surgically (detachable balloons may, in the future, permit achievement of this goal through percutaneous catheters), 2) to embolize the remaining minor feeders with percutaneous technique, and 3) ultimately to obliterate the site of the arteriovenous communication by directly injecting a rapidly polymerizing plastic. All three steps are essential; the ligation of major feeders and the embolic obliteration of minor ones would provide only temporary remission if the fistula itself is not occluded. Direct injection of embolizing plastic into the fistula, however, would increase the risk of pulmonary embolization without prior reduction of the high-flow condition.

Skinny or Chiba needles can be positioned, with relative safety, throughout the abdomen. Transit through bowel or solid viscera has not been associated with complications when No. 23 needles are used. We repeatedly punctured this pelvic arteriovenous fistula through an anterior abdominal approach, under fluoroscopic control using small injections of contrast medium for localization. This approach (the obstruc-
tion of major feeding arteries followed by the direct injection of the fistulous connection) provides an alternative therapeutic procedure when complete resection proves technically impossible. We have recently applied a similar technique to an unresectable congenital AVM with previously ligated feeding arteries.

References


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