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Giant Extramedullary Arteriovenous Fistula Supplied by the Anterior Spinal Artery: Treatment by Detachable Balloons

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A case of a large extramedullary arteriovenous fistula in a young man, which was deemed inoperable, is described. The fistula was mainly supplied by the anterior spinal artery and was responsible for a progressive myeloradicular syndrome. Successful treatment with detachable balloons was carried out. Along with this case report, a review is made of the different types of extramedullary arteriovenous fistulas and their treatment based upon 11 cases seen at Lariboisière Hospital: type 1 is a plain small fistula where embolization is contraindicated; type 2 shows an enlarged anterior spinal artery with dysplastic vein and may be embolized with solid particles; type 3 is a very large multipediculated fistula where the best treatment seems to be detachable balloons. The indications for using detachable balloons for temporary occlusion and for definitive treatment are reviewed. Special emphasis is placed on the possibility of detaching balloons without the need for a coaxial catheter.

Extramedullary arteriovenous fistulas fed by the anterior spinal artery were first described by Djindjian et al. [1] in 1977. Since that time we have documented 11 such cases at Lariboisière Hospital [2]. In one case in which surgical treatment was contraindicated the patient was treated with detachable balloons with considerable clinical improvement.

Case Report

A 34-year-old man had a subarachnoid hemorrhage in 1962 without localizing features. Carotid angiography revealed no abnormalities. In late 1980 he began to experience pain in his right knee and a burning sensation on the medial surface of his right thigh as well as spontaneous right patellar clonus. The patient also experienced frequency of micturition and exertional fatigue in his right leg.

In May 1981, the exertional fatigue in his right leg became worse, hindering his ability to walk up stairs. In September 1981, he began to experience the same symptoms in his left leg. At this time he consulted physicians at the Maggiore di Milano Hospital where myelography and medullary arteriography were performed. Myelography disclosed large vascular defects and arteriography disclosed a very large arteriovenous fistula supplied by the anterior spinal artery from the right T10 intercostal and left L1 lumbar arteries and by the posterior spinal arteries from the left and right L2 lumbar arteries. At the time of his hospitalization in November 1981 clinical examination in the neurology department revealed pyramidal signs in both lower limbs with exaggerated reflexes and ankle clonus, all with right-sided predominance. Sensory testing disclosed a zone of hypoesthesia for all types of stimulation at the distal part of the right leg up to just above the right knee, moderate hypoesthesia in the proximal area of the right leg, and dysesthesia of a burning nature on the medial aspect of the right thigh and left knee.

An arteriogram was obtained (fig. 1A). The first step in the procedure was to try to determinate the level of the arteriovenous shunt. Using the right femoral approach, a 7 French catheter was inserted up to the T10 level to permit the passage of a 3 French Fogarty catheter into the origin of the anterior spinal artery. This artery was then occluded, and, by means of a second catheter introduced through the left femoral artery, the other feeders were opacified, that is, left L1 and left and right L2. At first, the shunt was estimated to be at the T11 level (converging point of the two anterior spinal arteries), but ultimately it appeared to be at the L1 level where the posterior spinal arteries joined the malformation (figs. 1B–1D).

The Fogarty catheter was then replaced by a catheter with a detachable balloon (Granier, Paris) inserted into a coaxial catheter. However, the coaxial catheter could not progress farther than the origin of the enlarged anterior spinal artery. The balloon was then allowed to navigate the vessels entering the fistula and passed into the venous side of the shunt. It was then brought back very slowly to the precise level of the shunt and inflated. Clinical examination revealed that the patient's symptoms were improved. However, an attempt to detach the balloon provoked pain in the lumbar region and the balloon was therefore left in place without detachment. The next day, the patient began to move his legs and no longer had a motor deficit.

Four days later, a second spinal arteriogram was obtained. This showed that the balloon had deflated. At the same time, the patient began to deteriorate to the same clinical state that existed before therapy.

After another 3 weeks, arteriography and embolization were repeated. This time, we were able to detach a balloon at the level of L1; just beyond the shunt on the venous side. After detachment, it migrated up the posterior vein to T11 (fig. 2). Another balloon was then detached at the origin of the anterior spinal artery at the right T10 level and a third balloon at the origin of the left L1 feeder in order to decrease the blood pressure in the occluded segment (fig. 2C). This was possible since a normal supply to the anterior

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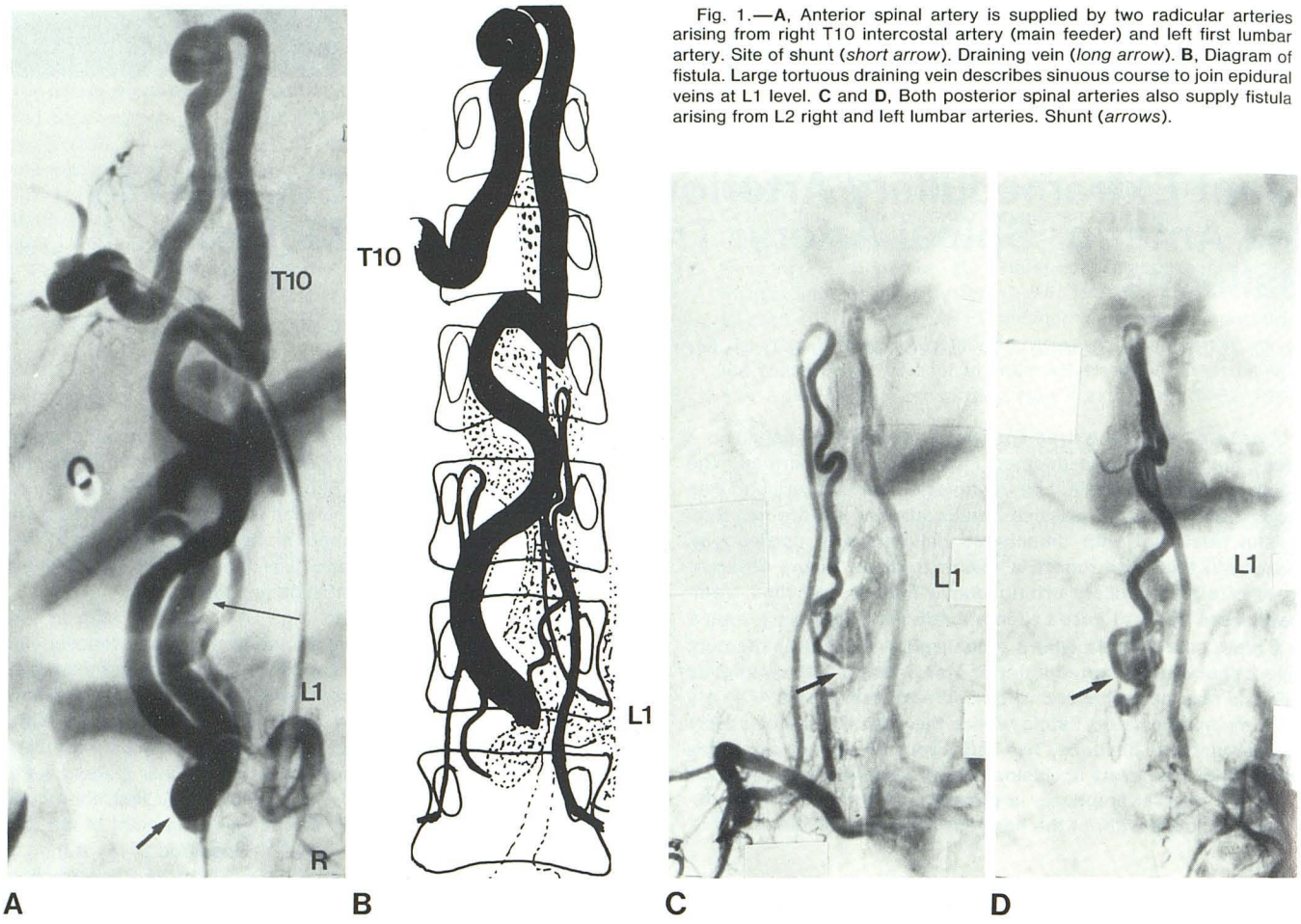


Fig. 1.—**A**, Anterior spinal artery is supplied by two radicular arteries arising from right T10 intercostal artery (main feeder) and left first lumbar artery. Site of shunt (*short arrow*). Draining vein (*long arrow*). **B**, Diagram of fistula. Large tortuous draining vein describes sinuous course to join epidural veins at L1 level. **C** and **D**, Both posterior spinal arteries also supply fistula arising from L2 right and left lumbar arteries. Shunt (*arrows*).

spinal artery on the upper segment was visualized. However, by such occluding of the two main inferior supplies to the anterior spinal artery including the artery of Adamkiewicz, after careful temporary occlusion test, we could demonstrate that there was a complete shunt to the fistula and these arteries were no longer functional for the spinal cord. The balloons were released by gentle traction and not with a coaxial catheter.

After release of the first balloon, the patient complained of lumbar pain similar to the first procedure. A follow-up angiogram showed complete disappearance of the malformation, even when opacifying the posterior spinal arteries at the left and right L2 levels. Two days after embolization, lumbar pain was observed and motor and sensory deficits were unchanged. Four days later, another spinal arteriogram was obtained, which showed that the two vessels remained well occluded and the balloons were still in place. Clinically the patient began to improve. Over the next week, recovery of motor function in his right leg stabilized. He was able to ride a bicycle, but still complained of a heavy feeling in the lumbar area both at rest and during exertion, and paresthesia in his right leg. On examination, there was decreased sensory perception to pain and temperature of a distal stocking-type distribution in the right leg. He also felt that he was walking on a cotton surface with his right foot. In addition there was continuing frequency of micturition and nocturia.

The patient was seen again in April 1982, 5 months after embolization. Spinal arteriography failed to opacify the fistula (fig. 3). He showed continued improvement. His gait was normal and he could run once again. Pyramidal signs persisted, but there were no objective sensory problems. He has returned to a normal life.

Discussion

This case is that of a young man with the onset of a slowly progressive asymmetric myeloradicular syndrome predominating in the right lower extremity. The syndrome appears to affect preferentially the territory of the anterior spinal artery with both motor weakness and sensory disturbances for pain and temperature.

Clinically, an arteriovenous fistula cannot be distinguished from any other type of medullary lesion. The history, however, of an unexplained subarachnoid hemorrhage 18 years before could indicate a vascular etiology. In fact, subarachnoid hemorrhage occurred in four of our 11 previously documented cases. It preceded the appearance of objective clinical findings by a long time (10–20 years), except for one case in which it occurred during the course of the active part of the illness.

In comparison with intramedullary arteriovenous malformations where subarachnoid hemorrhage and acute attacks occur frequently, the symptom-free interval is much longer. In both cases, however, the diagnosis is generally not made unless there are symptoms referable to a spinal medullary origin. In the case of this patient, carotid angiography was negative.

Clinically, it appears that once impairment has begun, a progres-

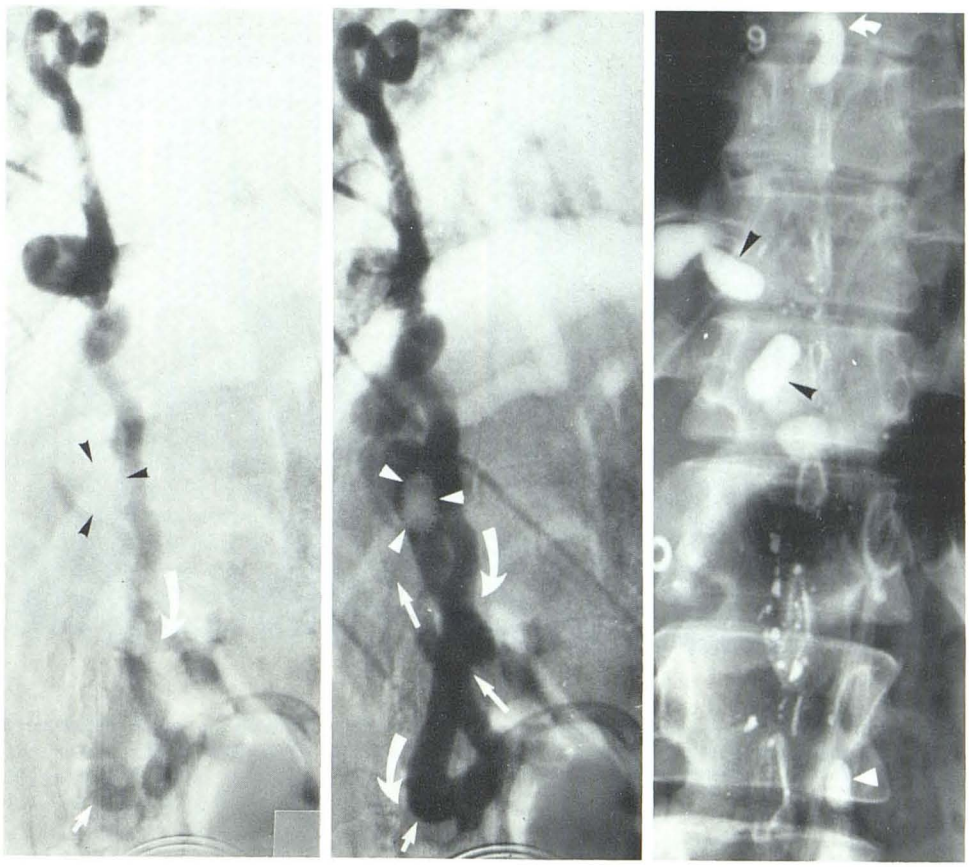


Fig. 2.—**A** and **B**, Lateral views. Course of balloon (*arrowheads*) after release may be followed beyond shunt (*short arrows*) in draining vein (*long arrows*) at posterior side of spinal cord. Artery (*curved arrows*). **C**, Plain film, anteroposterior view immediately after treatment. The three balloons (*arrowheads*) are in place and detached. Stagnation of contrast medium in superior part of anterior spinal artery (*arrow*) shows that occlusion is complete.

sive unremitting evolution follows that results ultimately in complete paraplegia. This is in contrast to intramedullary malformations, which in nearly 80% of the cases present as acute relapsing episodes [3].

In cases of meningeal extramedullary fistulas with medullary venous drainage [4], neurologic deficits also follow a progressive course, but their onset is usually later (between 50 and 70 years).

Neuroradiology

The diagnosis of a vascular disorder was initially made by myelography. Arteriography revealed the morphology of the malformation: a large direct "full-channel" extramedullary fistula with a shunt at the junction of the conus terminalis and cauda equina. Among the documented cases of extramedullary arteriovenous fistulas supplied by the anterior spinal artery (11 cases) at Lariboisière Hospital [2], we have distinguished three main types: plain fistula, with moderately enlarged, but no dysplastic vessels (three cases); enlarged vessels with a venous dysplastic aneurysm (four cases); and giant "full-channel" fistulas with dysplastic vessels (four cases). The exact site of the shunt may be difficult to locate if there is no difference in the caliber of the vessels. However, since the fistula often has multiple feeders, the point of convergence of the small-sized collateral feeders identifies the area of the shunt.

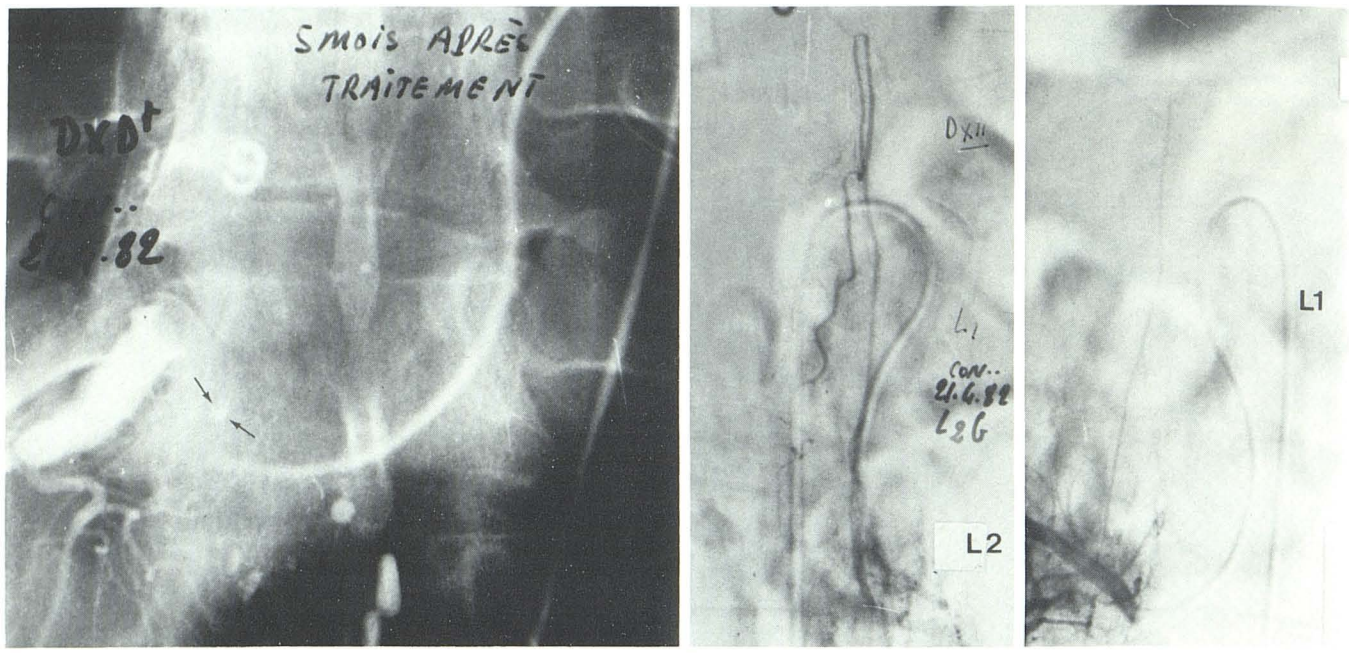
In our patient, we found one principal artery, the anterior spinal artery, was fed from the T10 level, with a smaller feeder below it at L1. Both posterior spinal arteries also supplied the malformation and they arose from the L2 level bilaterally.

Treatment

Treatment, surgical or by embolization, depends on the type of lesion. Small fistulas (type 1) situated at the level of the conus are supplied by one long thin anterior spinal artery often arising at a high level. The emboli have no chance of reaching the shunt and embolization may occlude the anterior spinal artery itself. This is therefore a good indication for surgery.

When the fistula is larger and the anterior spinal artery is long (type 2), a combined surgical and embolization plan may be considered. In one case, a balloon was positioned at the origin of the anterior spinal artery immediately before surgery and was then inflated when the surgeon reached the shunt after a small laminectomy. In other cases, treatment was with embolization alone using solid emboli.

With giant fistulas (type 3), surgery is impossible because of the enormous size of the vessels. Treatment by embolization is the only solution. Surgically opening the dura mater alone can provoke a cataclysmic hemorrhage (our unpublished case). We have success-



A B

Fig. 3.—**A**, Control arteriogram at T10 level 5 months after treatment. Balloon occluding origin of ASA is almost entirely deflated (*arrows*). Nevertheless, occlusion of ASA is still complete. **B**, Opacification of posterior spinal artery at L2 level bilaterally shows that these arteries have returned to normal caliber and fistula is no longer visible.

fully treated a similar case mainly supplied by a posterior spinal artery also using a detachable balloon.

Detachable Balloon Technique

Embolization with either solid or liquid material was impossible owing to the size of the shunt and the absence of an intermediate vascular network. Detachable balloons overcame this problem.

Temporary occlusion. After identifying the level of the shunt, the possibility of occluding the shunt was tested by leaving the balloon in place for 24 hr while observing the patient. A clinical improvement was noticed, indicating possible successful embolization. The test is important in patients with intramedullary malformations located at the upper dorsal level where it is known that the anterior spinal artery receives no collateral supply. Here, a temporary occluding test combined with clinical examination and opacification of the anterior spinal artery above (cervical) and below (dorsolumbar) help to determine whether embolization may be carried out safely.

Release without a coaxial catheter. Because of the sinuous nature of the anterior spinal artery in this case, it was obvious that a coaxial catheter could not be used. Since it could not go beyond the origin of the radicular artery at the T10 level feeding the anterior spinal artery, it was therefore necessary to try to detach the balloon by traction, after having inflated it sufficiently to affect the walls of the vessels. This traction of the balloon on the spinal cord and its vessel probably caused the lumbar pain in the patient during the first procedure. It was only by exerting a progressive and slow traction on the catheter that, on the second procedure, the balloon could be successfully detached without incident.

REFERENCES

1. Djindjian M, Djindjian R, Rey A, Hurth M, Houdart R. Intradural extra-medullary spinal arterio-venous malformation fed by the anterior spinal artery. *Surg Neurol* **1977**;8:85–94
2. Gueguen B. *Les fistules arterio-veineuses extra-medullaires alimentées par des artères à destinée médullaire* (thesis). Paris: Université de Paris VII Faculté de Médecine Lariboisière Saint-Louis, **1982**
3. Riché MC, Modenesi-Freitas J, Djindjian M, Merland JJ. Arterio-venous malformations of the spinal cord in children. *Neurology* (NY) **1982**;22:171–180
4. Merland JJ, Riché MC, Chiras J. Les fistules arterio-veineuses intra-canalaires extra-médullaires à drainage veineux médullaire. *J Neurol* **1980**;7:271–320