Combined Arteriovenous Malformation and Intramedullary Lipoma: Surgery and Embolotherapy

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Arteriovenous malformation (AVM) and intramedullary lipoma of the spinal cord are rare conditions [1-3]. To our knowledge, no report of these two lesions coexisting at the same level of the spine has been made. We treated such a case and are reporting it because of the diagnostic and therapeutic problems.

Case Report

A 23-year-old woman was first seen in the emergency room with a severe headache. She had awakened at home that day with a headache, walked to her mother's room, and passed out. On arrival at the hospital, she was lethargic but arousable and spoke only in monosyllables. Examination of the cranial nerves revealed a small hemorrhage below the right optic disk but no other abnormality. On motor examination, there was a slight atrophy and weakness of the left arm and leg that has been present since childhood. Sensory and cerebellar examinations were normal. There was marked nuchal rigidity, and a spinal puncture and CT scan showed evidence of a significant and diffuse subarachnoid hemorrhage. Cerebral angiography shortly after admission showed no intracranial aneurysm or AVM.

The history in this case is interesting and significant. A Wilm tumor was removed in infancy and radiation therapy was given to the left paraspinous area. The treated area extended from the level of the clavicle to the lower lumbar plexus, and hyperpigmentation of the skin was noticeable over this region. The left arm and left leg were first noted to be smaller than the right several years after the radiation. Severe hypertension, requiring four medications to control, had developed in early childhood and was attributed to a diffusely narrowed aorta, or pseudocoarctation, thought to be from the radiation therapy.

The patient's first hospital course was one of a slow but steady recovery over 3 weeks. The patient was discharged, but 4 weeks later was found to have developed a spastic paraparesis with a modified Brown-Séquard type of deficit. The headache and lethargy caused by her subarachnoid hemorrhage had completely resolved. She was readmitted for evaluation.

Metrizamide myelography was performed with a CT scan using the lateral C1-C2 approach (fig. 1). An intradural extramedullary mass lesion displaced the cord to the right and extended from C7 to T1-T2. Both the CT scan and myelography showed, in addition to the smooth oval mass, discrete curvilinear defects in the column of contrast material that were thought to be enlarged blood vessels. A large component of fat was identified in the mass by the CT scan, indicating the presence of a lipoma [4]. Because of the previous subarachnoid hemorrhage without an obvious source and the suspicious vascular structures at myelography, spinal angiography was performed (fig. 2). This showed an AVM supplied by three major arterial feeders, all arising from the left costocervical trunk and entering the spinal canal at T1-T2. The normal anterior spinal artery at this level arose from the lower part of the right vertebral artery. Upper thoracic segmental branches showed slight collateral filling of these principal feeders to the malformation but no direct filling of the malformation. Dense thickened bone was also evident on the left side at these spinal segments.

The patient's weakness progressed in the hospital. After 4 days, she could barely lift the left thigh against gravity in bed; the right leg could be moved through a full range of motion but was markedly weak. The long-standing weakness of the left arm and hand was unchanged. Analgesia was present over much of the right leg; moderately severe hypalgesia was present over the left leg; and truncal analgesia was present from T3 to T6.

Because of neurologic deterioration, a C7-T3 laminectomy was performed. An intramedullary lipoma was found as predicted from the neuroradiologic studies. The smooth exophytic component of the mass was removed, but the lipoma entered the spinal cord at the same level at which the AVM was located. Enlarged vessels were seen bulging through the pia from inside the enlarged spinal cord in several locations, but the nidus of the malformation was not found on the surface of the spinal cord. Considerable arachnoid adhesions were present, presumably from the previous radiation. Because of these findings, a limited removal of the intramedullary portion of the lipoma was made, and the AVM, which was thought to be intramedullary and probably within the lipoma as well, was not removed. A wide laminectomy was done, and the dura was left open and covered with Gelfoam to decompress the enlarged spinal cord.

The patient's postoperative course was at first benign. After a slight deterioration in strength, she began to improve and displayed increased strength and range of movement in both legs relative to her preoperative state. At about 1 week after surgery, however, the weakness returned and she was also occasionally incontinent of urine. Her neurologic disability continued to increase, and she was readmitted.

Repeat angiography with oblique and lateral views showed the malformation lying posterolaterally in the canal on the right side.
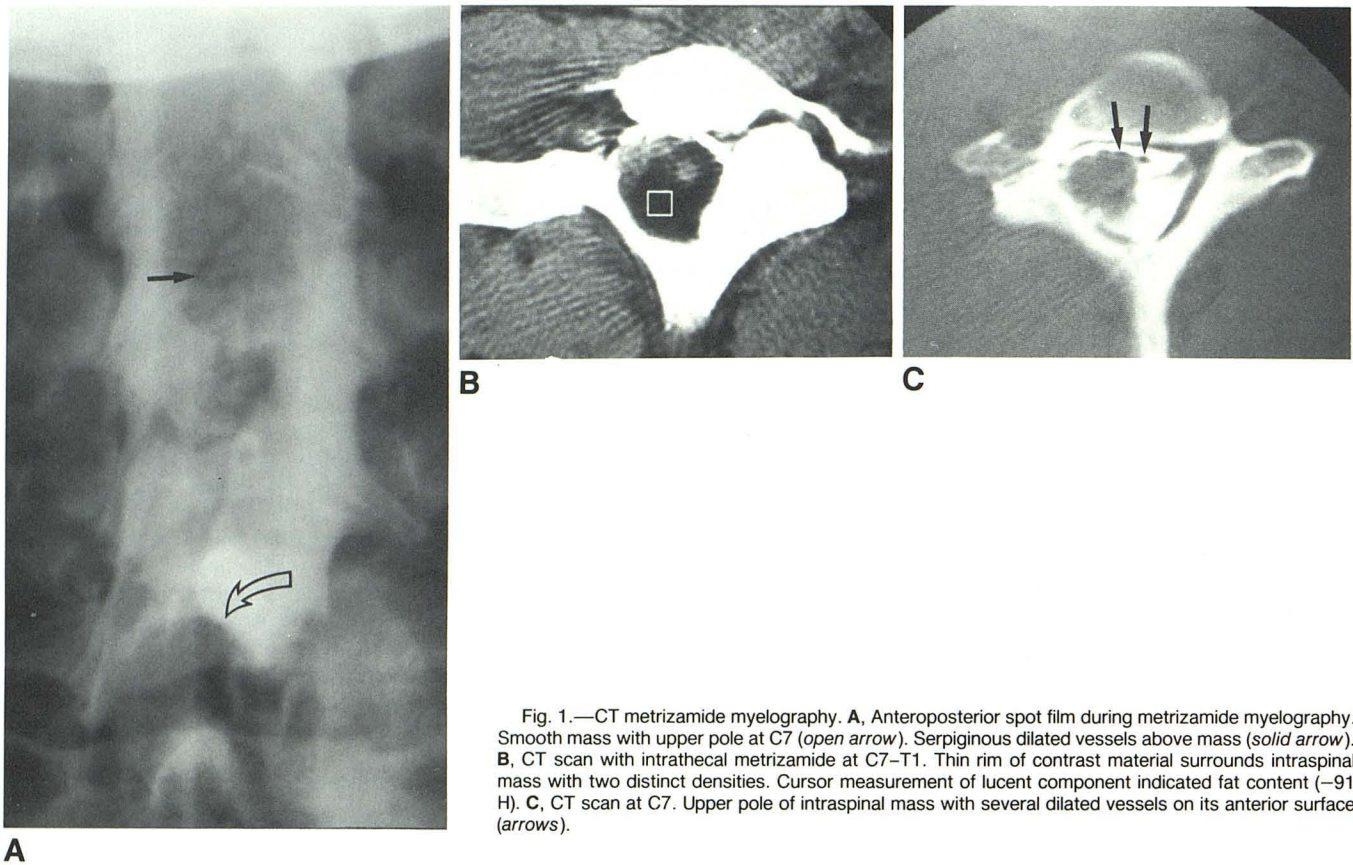
A

B

C

Fig. 1.—CT metrizamide myelography. **A,** Anteroposterior spot film during metrizamide myelography. Smooth mass with upper pole at C7 (*open arrow*). Serpiginous dilated vessels above mass (*solid arrow*). **B,** CT scan with intrathecal metrizamide at C7–T1. Thin rim of contrast material surrounds intraspinal mass with two distinct densities. Cursor measurement of lucent component indicated fat content (−91 H). **C,** CT scan at C7. Upper pole of intraspinal mass with several dilated vessels on its anterior surface (*arrows*).

Embolization of the malformation was performed with a 5 French catheter and a slurry of granular polyvinyl alcohol (PVA) foam particles (Unipoint Industries, High Point, NC) measuring 250–590 µm suspended in normal saline [5]. Small increments of the embolic material were injected slowly with contrast material under fluoroscopic control. A venous aneurysm arising from the nidus was thought to be the source of hemorrhage, and priority was given to eliminating that part of the malformation. The three feeding vessels, including the vessel supplying the aneurysm, were successfully occluded (fig. 3).

The patient's neurologic condition worsened after embolization. At 12 hr postembolization she had only trace movement of the toes and foot on the right and no movement of the left leg or foot, along with increased hypalgesia. She remained in the hospital for 2 weeks, during which time her motor and sensory function began to improve.

Physical therapy was begun, and over the next few months a marked neurologic improvement occurred. By 6 months after embolization, the patient was able to walk with a long leg brace on the left leg, no brace on the right leg, and the walker; she no longer required catheterization of her bladder. Her neurologic function had improved relative to its preembolization status. Repeat angiography showed virtually complete obliteration of the AVM nidus. Because of her continued improvement, it was decided not to embolize further, but to recheck the lesion in 2–3 years and reconsider embolization if any malformation was still present.

She continued to improve in the year since embolization and was able to walk alone with a cane and no leg braces. She was experiencing similar resolution of her sensory deficits.

Discussion

The patient had two serious neurologic problems: (1) spinal subarachnoid hemorrhage, with its attendant long-range problems and the additional morbidity of further hemorrhages, and (2) progressive myelopathy presumably from the mass effect of the intradural lipoma or vascular changes in the spinal cord from the AVM.

Therapy of the lipoma was undertaken first, as the compression myelopathic changes were the most serious problem facing the patient. Surgical decompression effected only a short-term improvement. The cause of her postoperative deterioration is unknown. Microvascular alterations caused by partial surgical excision of the lipoma may have impaired venous drainage of the malformation or increased the steal phenomenon from normal cord tissue.

Embolization was undertaken with two goals in mind: to diminish the threat of further hemorrhage and to improve the perfusion of the spinal cord by diminishing the steal or venous congestion secondary to the malformation [6].

The patient's acute neurologic deterioration after embolization was probably due to edema either in the spinal cord or the lipoma. The almost complete return of cord function indicates that very little infarction of cord tissue occurred. Angiographic studies indicated that the three principal feeders
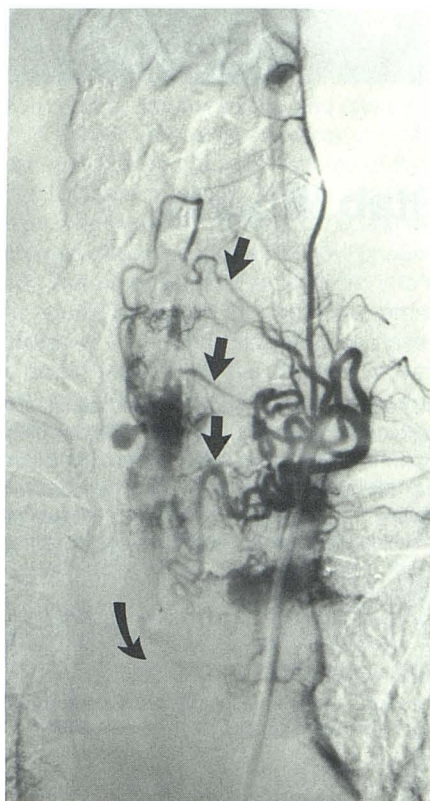
Fig. 2.—Spinal angiogram, selective left costo-cervical trunk injection. Intraspinal AVM with three main feeding vessels (*straight arrows*). Venous aneurysm projects laterally. Veins drain inferiorly (*curved arrow*).
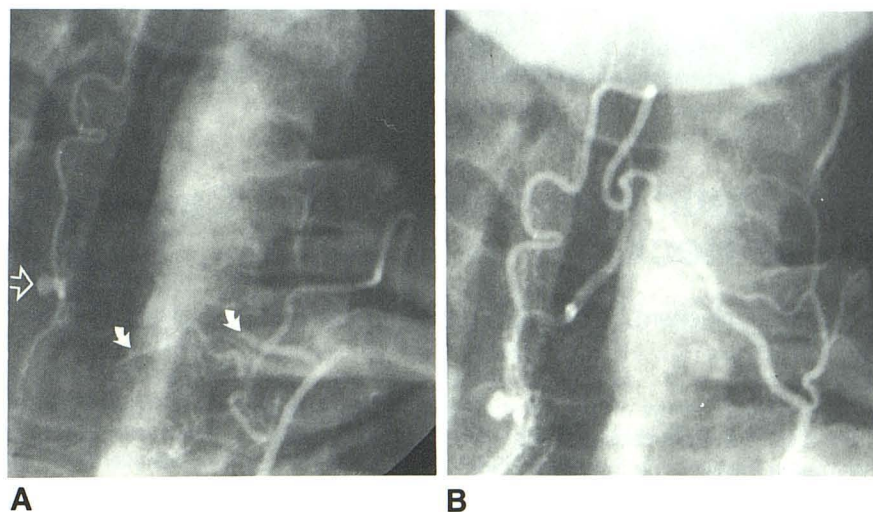

Fig. 3.—Embolization. **A**, Two lower feeding vessels have been occluded (*solid arrows*). Uppermost vessel remains, supplying area of malformation including venous aneurysm (*open arrow*). **B**, Repositioning catheter subselectively in uppermost vessel shows detail of malformation, aneurysm, and venous drainage. This vessel was then occluded with particulate PVA foam.

supplied the dorsolateral aspect of the cord and that the anterior spinal artery was not involved in the malformation, providing some margin of safety for the planned embolization. The coexistence of the lipoma with resultant distortion of the cord within the neural canal did introduce some uncertainty regarding the orientation of the malformation nidus relative to major cord structures, however.

In this case finely granular PVA foam particles were used for embolization. This material is easily introduced through small catheters and its occlusive features are permanent and are easily monitored angiographically [5]. Liquid embolic materials, either silicon rubber or cyanoacrylics, likely would have provided similar results.

The intermingling of the malformation within the lipoma made the surgical removal of both lesions an unacceptable hazard. However, partial removal of the lipoma with decompression laminectomy followed by embolization of the AVM produced a remarkable improvement in this patient who was completely bedridden and who can now walk.

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