Neurosurg 71:768-771, 1989

Sacral lipoma of the filum terminale with dural arteriovenous fistula

Case report

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A patient presenting with progressive paraparesis was found to have a dural sacral arteriovenous (AV) fistula. His condition deteriorated abruptly after thoracolumbar angiography. Embolization of the fistula improved the patient's status so that he was able to walk with crutches. One year later his neurological condition worsened. He was treated via an enlarged laminectomy because of uncertainty concerning a lipoma noted on the initial computerized tomography scan. The lesion consisted of an intradural filum terminale lipoma associated with an AV fistula, both of which were excised. The patient's condition was unchanged 6 months later. The different types of spinal lipomas and spinal AV malformations are reviewed, and mechanisms are proposed to explain the clinical deterioration in this patient. Venous hypertension seems to be the most likely possibility. The lipoma may have produced local hypervascularization of the dura mater with a subsequently acquired AV fistula.

KEY WORDS • filum terminale • spinal neoplasm • lipoma • arteriovenous fistula • dural fistula

Spinal lipomas are rare, with an overall incidence approaching 1% of all spinal axis tumors. They are usually considered to consist of three subtypes: 1) intramedullary lipomas, 2) lipomyelo(meningo)celes affecting the spinal cord, and 3) lipomas of the filum terminale with limited involvement of the lowest level of the conus medullaris. Spinal arteriovenous malformations (AVM's) are also uncommon and are best classified by angiography. Intramedullary intradural AVM's are of congenital origin with symptoms occurring before the age of 40 years. On the other hand, dural arteriovenous (AV) fistulas are considered to be acquired lesions, encountered in patients aged over 40 years. Sacral dural AV fistulas are exceptional; only seven such cases have been found in the literature. To our knowledge, only one case of an AVM and a lipoma coexisting at the same level has been reported: a combined intramedullary AVM and lipoma at the cervicothoracic level.

The present case report describes a lipoma of the filum terminale associated with a sacral dural AV fistula. We have reported it because of the diagnostic, pathogenic, and neurological problems involved.

Case Report

This 53-year-old man presented with a 6-month history of neurological claudication with dysuria. The symptoms were temporarily intensified by exercise and ventral compression during a computerized tomography (CT) scan for kidney exploration. No relevant personal or family medical history was elicited.

First Admission. Neurological examination was normal except for the absence of deep-tendon reflexes in both legs. Metrizamide myelography, performed by the lumbar route, revealed serpentine vessels; however, thoracolumbar angiography failed to demonstrate any AVM or AV fistula. After thoracolumbar angiography, the patient was found to have complete flaccid paraplegia with anesthesia below T-12 and bilateral Babinski signs. Partial recovery was seen during the 1st week after the examination. Repeat myelography with CT scanning showed serpentine vessels within the sacral canal and a moderately large mass, initially considered to be a dilated pseudotumoral vein although its density was compatible with a lipoma. Bilateral hypogastric angiography demonstrated a dural AV fistula fed by the right lateral sacral artery and draining into the premed-
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FIG. 1. Angiograms (upper) and schematic representation (lower) of the lesions. AVF = arteriovenous fistula (broken line). Left Pair: Selective arteriography, arterial and venous phases, of the right lateral sacral artery (RLSA) via the hypogastric artery (C = catheter). A dilated vein of the filum terminale (large arrow) was rapidly injected by retrosacral anastomosis (small arrows, lower). There was also injection of the median sacral artery (MSA) and left lateral sacral artery (LLSA). Center Pair: The dural AVF (arrows) was fed by two dural arteries (arrows). Right Pair: The AVF (arrows) drains via the ascending vein of the filum terminale into the premedullary veins (arrow).

ullary veins via the filum terminale vein (Fig. 1). The AV fistula was treated by embolization and was occluded. The patient's neurological condition continued to improve, and after a month he was able to walk alone with two canes, although he had partial sensory deficits below L-4.

Second Admission. One year after embolization, the weakness returned and the patient was readmitted. Laminectomy extending from L-5 to the sacral canal was performed without a repeat angiography. A lipoma was found attached to the filum terminale and to the "cul de sac" of the dura mater, which appeared very
vascular. An enlarged vein of the filum terminale was seen embedded in the lipoma (Fig. 2). Complete resection of the lipoma and section of its draining vein with the filum terminale was performed. The tumor and filum terminale were sent to the pathology department for examination (Fig. 3). Six months after the operation, the patient’s condition was unchanged.

Pathological Findings. Histological examination of the surgical specimen showed a mass of white adipose tissue containing numerous disseminated sections of small fascicles of myelinated and unmyelinated peripheral nerve fibers (one of them containing a few neuronal perikarya). In several sections, small arteries, arterioles, venules, veins, and capillaries were seen. Between the adipocytes, a delicate network of reticulin fibers was present. A dense capsule of collagenous fibrous tissue with small blood vessels and fascicles of peripheral nerve fibers surrounded the lesion. At the periphery of the mass, within the capsule, a small group of ependymal cells was identified; adjacent to it was found a voluminous vascular pedicle composed of a section of a muscular artery and its satellite vein. The vein was of large caliber and its thickened wall showed some histological signs of arterialization. The ultrastructural study confirmed the nature of the various components identified by light microscopy. In fact, the pathological mass looked like a normal filum terminale filled by an abundant proliferation of adipose tissue and would be more accurately described as a lipomatous filum than a lipoma of the filum. Histologically, there was no morphological evidence of the dural AV fistula shown on angiography. No vascular malformation or any other type of abnormal blood vessel was seen.

Discussion

Vascular filling defects at myelography are not pathognomonic. They may be seen associated with AVM’s of various types with certain intramedullary or cauda equina tumors, particularly hemangioblastoma, ependymoma, or combined AVM and lipoma. These tumors can be revealed by myelography and CT scanning, but interpretation may be difficult. Magnetic resonance imaging seems to be of more help.

The patient’s progressive symptoms could have been due to three possible mechanisms. The symptoms and signs may have been caused by the mass effect from the tumor, but they cannot be explained by the level of the lipoma. A second possibility is tethering of the cord because of the lipoma; in fact, on CT scanning with metrizamide the conus medullaris was seen at the L-3 vertebral level. The third possible cause of symptoms was chronic venous hypertension from the AV fistula. Angiography of an ischemic spinal cord due to tethered cord or venous hypertension can cause abrupt exacerbation of symptoms. Venous hypertension seems to be the most likely pathogenic mechanism. Aggravation after ventral compression during CT exploration is another argument in favor of this possibility.

Embolization was undertaken to improve perfusion of the conus by reducing the venous congestion secondary to the AV fistula. Angiographic findings were typical of a dural AV fistula and therefore supported the diagnosis of the combination of an AV fistula with

Fig. 2. Operative view, posterior approach. The tumor (T) is attached to the filum terminale and to the “cul de sac” of the dura and the dilated vein of the filum terminale (V).

Fig. 3. Cross sections of the lipoma and the enlarged filum terminale vein.
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lipoma of the filum terminale. A lipoma fixed to the "cul de sac" of the dura mater (a site of potential AV communications)\textsuperscript{1} may have produced local hypervascularization with a subsequently acquired AV fistula. Among the various etiologies of acquired dural AV fistula, one must consider the possibility of a tumor as in this case.

References

Manuscript received January 10, 1989.
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