

CAVERNOUS ANGIOMA OF THE CAUDA EQUINA

Case Report

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ABSTRACT - We present a rare case of cavernous angioma of the cauda equina and review the eleven cases available in the literature. A 44-year-old woman presented with low back pain and sciatica associated with bowel and bladder dysfunction and motor weakness of the lower extremity. The MRI revealed an enhancing, heterogeneous and hyperintense intradural lesion compressing the cauda equina roots at the L4 level. Laminectomy at L3-L4 and total removal of the tumor were performed without additional neurological deficit. Pathology revealed a cavernous angioma. The literature, clinical presentation, technical examinations, and treatment are reviewed.

KEY WORDS: cavernous angioma, cauda equina, cavernoma.

Angioma cavernoso de cauda equina: relato de caso

RESUMO - Relatamos um caso de angioma cavernoso de cauda equina em mulher de 44 anos de idade com sintomas de lombocatalgia associada a fraqueza de membros inferiores e disfunção esfinteriana vesical e anal. Exame de ressonância magnética evidenciou lesão expansiva intradural heterogênea e hiperintensa na cauda equina. Indicado tratamento cirúrgico com remoção completa através de laminectomia L3 e L4. O exame anatomopatológico foi compatível com angioma cavernoso. Os onze casos encontrados na literatura são revisados correlacionando a apresentação clínica, tratamento proposto e prognóstico.

PALAVRAS-CHAVE: angioma cavernoso, cauda equina, cavernoma.

Cavernous angioma is a vascular malformation created by anomalous vessels without interposition of neural tissue¹. Primary tumors of the filum terminale and conus medullaris, or both, comprise only 6% of spinal cord tumors². Only 6.2% to 7.5% of all intraspinal tumors are vascular^{3,4}. The cavernous angioma rarely occurs in the cauda equina. The literature reports eleven cases⁵⁻¹⁴.

We report on a woman with a cavernous angioma of the cauda equina presenting with low back pain with sciatica, motor and sensory deficit of the lower extremities and bladder and bowel sphincter dysfunction.

CASE

A 44-year-old woman presented with a 4-month history of low back pain and bilateral sciatica. The pain was exacerbated by ambulation. The pain became worse in the last 10 days and was associated with lower extremity numbness, difficulty in walking and bladder and bowel control sphincter loss. The neurologic examination demonstrated bilateral loss of pin prick at L4, L5 and S1 dermatomes,

right absence of Achilles reflex, bilateral positive straight leg raising test and a paresis during the examination of plantar flexion and dorsiflexion of the right foot. The urinary postvoid residual was 200-mL. Magnetic resonance imaging (MRI) of the lumbar spine showed a 30 mm x 20 mm heterogeneous enhancing and high signal intensity mass completely obliterating the spinal canal at the L4 level (Fig 1). High-dose steroids was started and surgery was indicated.

After a laminectomy at L3 and L4 the dura was tense and without pulsation. The dura and arachnoid were opened under microscopic magnification. A 3-cm dark-bluish tumor was seen, displacing the caudal nerve roots circumferentially around it. After separation from the arachnoid membranes, the tumor still adhered closely to a thin root that could not be dissected free at the tumor center because it disappeared inside the capsule (Fig 2 and Fig 3).

After cutting the root, the neoplasm was totally removed. Histopathological examination revealed many narrow vessels and large dilated vascular channels totally or partly obstructed by thrombi. The diagnosis was cavernous angioma (Fig 3). The postoperative course was uneventful, and the patient was discharged after 6 days without pain, the bladder and bowel function returned to normal, absence

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of the right Achilles reflex, lower extremity paresthesias and the motor strength of the right foot was almost normal. At the 6-month follow-up neurologic examination, no residual motor deficit of the right foot, lower extremity paresthesias and diminished right ankle reflexes were observed. Informed, free and written consent was signed by the patient for the publication her case.

DISCUSSION

Cavernous angioma represents 5 to 16% of spinal vascular anomalies preferably located at the vertebral bodies¹⁵. Cavernous angioma occurring within the cauda equina are even more rare, with only eleven cases reported in the literature⁵⁻¹⁴. Sometimes the cavernous angioma can be located at the lumbar epidural space¹⁶.

In these cases, eight of the eleven patients were male and three female, presenting between the ages of 20 and 67 years, 42.9 years being the average (Table 1). The clinical symptoms were related to subarachnoid hemorrhage (headache, nuchal rigidity and vomiting) in three cases^{7,10,11}, local compression of adjacent nerve roots (low back pain and sciatica, sensitive and motor disturbance of the lower limbs, sphincter dysfunction) in six^{5,6,8,11-14}, and, in two cases, symptoms of intracranial hypertension caused by hydrocephalus^{9,12}. The symptoms usually had acute onset in the subarachnoid hemorrhage patients, in the hydrocephalus patients the symptoms started four and three months before admission, and when the symptoms were related with expansive process, the average of beginning of symptoms ranged between ten days and nine years. In all previously reported cases, as in ours, the tumor was totally removed despite their close adherence to the nerve roots.

In the case described by Pagni et al.⁸, and our case, the tumor was excised with the attached thin nerve root because the root merging into the tumor made it impossible to individualize despite microsurgical dissection. This was seen in the



Fig 1. Preoperative T1-weighted MRI scan after gadolinium injection, showing heterogeneous hyperintense intradural cavernous angiomas at L4 level.

histological sections of the nerve root and tumor interface (Fig 3). Nevertheless, resection of this thin nerve root that becomes part of the tumor stroma did not compromise good recovery, probably because it was nonfunctional. In all cases, as in ours, there were gradual clinical improvements after lesion resection. In the cases presented with hydrocephalus there was an

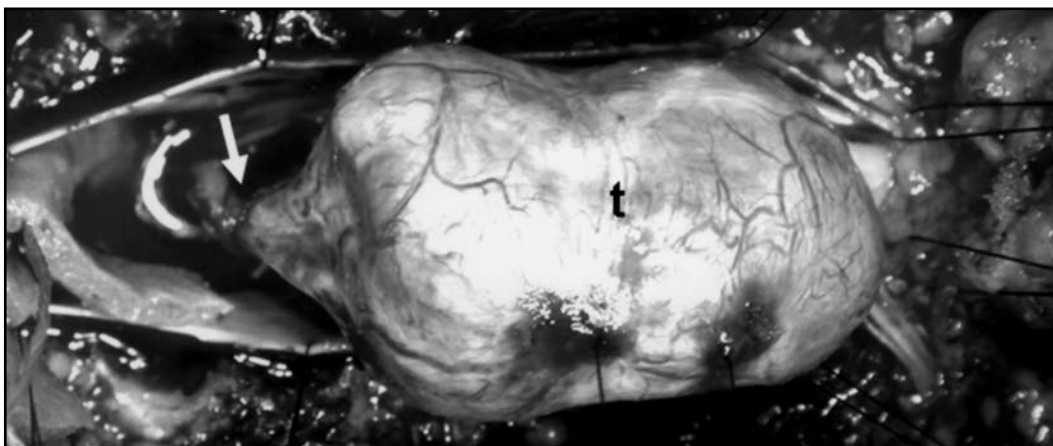


Fig 2. Intraoperative image showing the 30 mm x 20 mm berry-like tumor (t) after separation from the roots by blunt dissection. A rootlet remains adherent to the tumor disappearing inside its capsule (arrow).

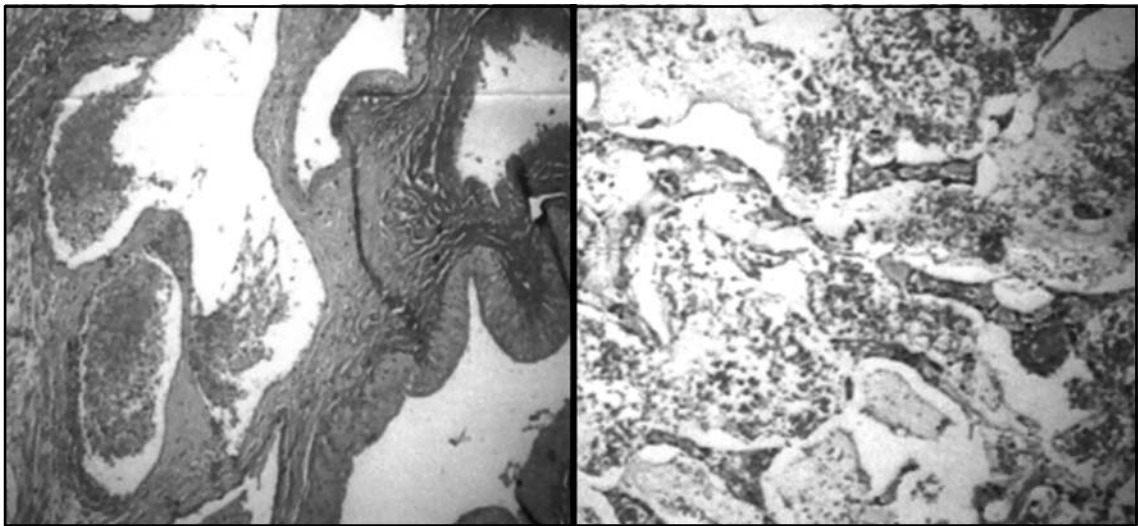


Fig 3. Left: Photomicrograph of the surgical specimen showing connective tissue with hemosiderine pigment and dilated vessels (H & H, x 100). Right: Photomicrograph showing the transition between rootlet and cavernoma, with gradually loss of cleavage planes between the nerve root and the tumor (H & H, x 100).

Table 1. Reported cases of cavernous angioma of the cauda equina.

Author (Year)	Age/ Gender	Presenting symptoms	Follow-up	Residual deficit
Hirsch et al. (1965) ⁵	20/ M	Pain, paraplegia, sphincter dysfunction, anesthesia, probable episode of subarachnoid hemorrhage	2 years	Right foot paresis, left sacral sensory deficit
Pansini et al. (1966) ⁶	46/ M	Pain, sphincteric/erectile dysfunction, anesthesia, right quadriceps atrophy	1 year	Erectile dysfunction
Ueda et al. (1987) ⁷	28/ M	Pain, subarachnoid hemorrhage with meningeal signs	3 weeks	None
Pagni et al. (1990) ⁸	46/ M	Pain, diminished Achilles reflex	20 months	Right lower extremity paresthesias
Ramos et al. (1991) ⁹	67/ F	Cognitive dysfunction, gait unsteady, sphincteric dysfunction	3 years	None
Bruni et al. (1994) ¹⁰	28/ M	Pain, subarachnoid hemorrhage with meningeal signs	7 days	None
Cervoni et al. (1995) ¹¹	26/ F	Pain, subarachnoid hemorrhage with meningeal signs	Discharge from hospital	None
Cervoni et al. (1995) ¹¹	32/ M	Deficit of the plantar flexion, anesthesia	6 months	Left plantar flexion, left L5 sensory deficit
Makino et al. (1995) ¹²	67/ M	Headache, gait unsteady	6 months	None
Moreno et al. (1995) ¹³	63/ M	Pain, paraplegia, anesthesia, sphincter dysfunction, absence patellar and ankle reflex	Not mentioned	Lower extremity weakness, pain, sphincter dysfunction
Duke et al. (1998) ¹⁴	49/ F	Pain, absence right Achilles reflex, anesthesia, sphincter dysfunction	3 months	Right S1 sensory deficit, absent right ankle reflex
Present report	44/ F	Pain, absence right ankle reflex, right foot paresis, anesthesia, sphincter dysfunction	6 months	lower extremity paresthesias and diminished right Achilles reflex

M, male; F, female.

improvement of the ventriculomegaly without shunt six months¹² and three years⁹ after tumor removal. These show that complete resection can be achieved safely using careful microsurgical technique. Residual deficit observed in the reported cases at the last follow-up was sexual impotence, foot paresis, diminished Achilles reflex and lower extremity sensory loss^{5,6,8,11-14}.

In conclusion, cavernous angiomas of the cauda equina are extremely rare lesions that may present as low back pain with sciatica, neurologic deficit, or as subarachnoid hemorrhage. They can be successfully treated by surgery.

REFERENCES

1. Simard J, Garcia-Bengochea F, Ballinger W, Mickle J, Quisling R. Cavernous angioma: a review of 126 collected and 12 new cases. *Neurosurgery* 1986;18:162-172.
2. Norstrom C, Kernohan J, Love J. One hundred primary caudal tumors. *JAMA* 1961;178:1071-1077.
3. Rasmussen T, Kernohan J, Adson A. Pathologic classification, with surgical consideration, of intraspinal tumors. *Ann Surg* 1940;111:513-530.
4. Costa LB Jr, Andrade A, Braga BP, Ribeiro CA. Cauda equina hemangioblastoma: case report. *Arq Neuropsiquiatr* 2003;61:456-458.
5. Hirsch J, Pradat P, David M. Angiome cavernoux de la queue de cheval. *Neurochirurgie* 1965;11:323-327.
6. Pansini A, Lo Re F. Raro case di angiocavernoma della cauda. *Men Soc Tos Um Chir* 1966;27:679-696.
7. Ueda S, Saito A, Inomori S. Cavernous angioma of the cauda equina producing subarachnoid hemorrhage. *J Neurosurg* 1987;66:134-136.
8. Pagni A, Canavero S, Forni M. Report of a cavernoma of the cauda equina and review of the literature. *Surg Neurol* 1990;33:124-131.
9. Ramos FJ, De Toffol B, Aesch B, Jan M. Hydrocephalus and cavernoma of the cauda equina. *Neurosurgery* 1990;27:139-142.
10. Bruni P, Massari A, Greco R, Hernandez R, Oddi G, Chiappetta F. Subarachnoid hemorrhage from cavernous angioma of the cauda equina: case report. *Surg Neurol* 1994;41:226-229.
11. Cervoni L, Celli P, Gagliardi F. Cavernous angioma of the cauda equina: report of two cases and review of the literature. *Neurosurg Rev* 1995;18:281-283.
12. Makino K, Takamura H, Gotoh S, Andoh M. Cauda equina cavernous hemangioma associated with hydrocephalus: case report. *No To Shinkei* 1995;47:783-787.
13. Moreno R, Romero J, Serrano V, Madrid A, Jarrín S, Casado J. Cavernoma intradural extramedular de cola de caballo. *Rev Neurol* 1995;23:1228-1230.
14. Duke B, Levy A, Lillehei K. Cavernous angiomas of the cauda equina: case report and review of the literature. *Surg Neurol* 1998;50:442-445.
15. Newton TM, Potts D. Computed tomography of the spine and spinal cord. In: *Vascular malformations*. San Anselmo, CA:Clavadel Press, 1983:398.
16. Félix A, Koerbel A, Hanel RA, Cichon E, Araújo JC. Angioma cavernoso epidural: relato de caso. *Arq Neuropsiquiatr* 2001;59:440-443.