

Hemorrhagic Schwannoma of the Cauda Equina: Case Report and Review of the Literature

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Abstract

Spinal intradural hemorrhage is a rare event; the most common causes of spinal bleeding are traumas, medical therapy with anticoagulants and thrombolytics, vascular malformations, and congenital defects of coagulation. Rarely, spinal cord tumors may cause hemorrhage. Herein, we report the case of a patient with acute and quickly worsening lumbar pain: the neurological examination revealed a flaccid paraplegia caused by an intradural lesion extending on the right side of the spinal cord from T1 to L2 vertebral level. Pathological examination revealed an hemorrhagic schwannoma. Acute spinal subdural hemorrhage caused by spinal schwannomas is a very rare occurrence (29 cases only have been previously reported). Review of the literature with clinico-diagnostic features is presented, surgical treatment is explained, and pathological findings with possible etiopathogenesis of hemorrhage are described.

Keywords: spinal bleeding, hemorrhagic schwannoma, intradural schwannoma, flaccid paraplegia

Introduction

Schwannomas are benign, typically encapsulated, nerve sheath tumors, composed entirely of well-differentiated Schwann cells.¹ They arise in the transition zone between central and peripheral myelination of nerve sheath, displacing or invading the normal structure of a nerve trunk. Schwannomas are most often located in the head and neck region, or along the limbs. Spinal schwannomas usually present as intraspinal lesions (accounting for 30%

of primary spinal tumor).² The most common presenting symptoms are progressive lumbar pain and radiculopathy, or chronic neurological deficits. Rarely, they may present with symptoms due to hydrocephalus.³ On very rare occasions, spinal schwannomas may cause subdural hemorrhage presenting with acute neurological deterioration. The purpose of this paper is to report a case of schwannoma of the cauda equina presenting with acute neurological dysfunction due to massive hemorrhage.

Case Report

A 57-year-old female with a past medical history of hypertension and obesity presented to an initial medical observation with acute and quickly worsening lumbar pain, associated with right lower limb weakness and bilateral impaired sensitivity with paraesthesia. There was no history of trauma or

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anticoagulation therapy. Five days after the onset of symptoms, the patient was referred to our Institution for a better diagnostic scrutiny, due to the rapid deterioration of clinical conditions. She was confused, agitated, and the neurological examination revealed a flaccid paraplegia with sensitive level around T11 dermatome and fecal and urinary

incontinence. The patient was submitted to a spinal magnetic resonance imaging (MRI) that showed an intradural lesion extending to the right side of the spinal cord from T1 to L2 vertebral level. The lesion appeared heterogeneously hyperintense on T2-weighted and hypointense on T2*-weighted gradient echo; the mass demonstrated patchy enhancement after

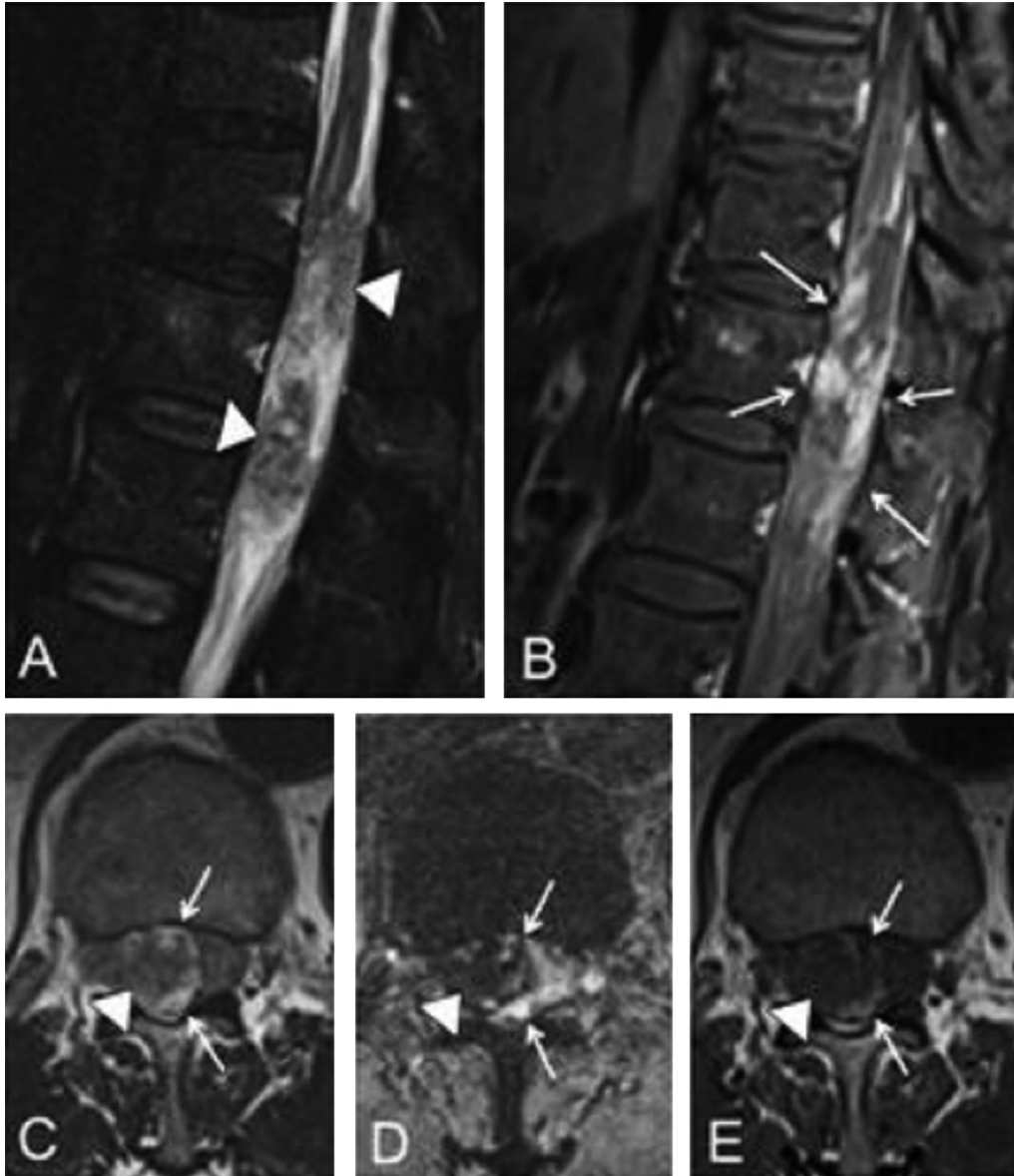


Fig. 1 Preoperative MRI. (A) Sagittal STIR T2-weighted and (B) sagittal T1-weighted post-gadolinium sequences showing an intra-dural extramedullary mass displacing anteriorly the conus medullaris. The lesion appears inhomogeneously hyperintense in STIR T2, with areas of reduced signal representing hemorrhagic components (arrowhead in A), and patchy enhancement after contrast medium administration (white arrows in B). (C) Axial T2-weighted, (D) axial T2*-weighted and E axial T1-weighted post-gadolinium sequences show the mass enlarging the vertebral canal and shifting on the left side the spinal cord. The hemorrhagic portion (arrowhead) is better depicted in the sequence susceptible to paramagnetic effect (more evident loss of signal due to deoxyhemoglobin in D), and is surrounded by a thin area of T2 hyperintensity (white arrows in C and D) with patchy contrast enhancement (white arrows in E). MRI: magnetic resonance imaging, STIR: short time inversion recovery.

contrast medium administration (Figs. 1A–1E). The lesion was considered intradural and extramedullary since it caused a left ward deviation of the spinal cord. Moreover, the diameter of the spinal canal was slightly increased at T12 level, suggesting the slow growing nature of the mass.

Bleeding due to intradural extramedullary hemorrhagic neoplasm was proposed.

Due to the severe neurological conditions, surgical therapy was delivered as an emergency. The patient was submitted in less than 24 hours to a decompressive T11-L1 laminectomy, exposing the dural sac (Figs. 2A–2C), which was extremely swollen, with ectatic vessels around. The dura was opened applying microsurgical technique and a large

brownish mass was detected inside the roots of the cauda equina (Fig. 2D). The mass was attached to an ectatic vessel perforating the dura mater. A large hematoma was present below the lesion, enveloping the nervous structures. After careful removal of hemorrhagic clots around the roots of the cauda equina, the mass was totally resected detaching it from the abnormal neurovascular pedicle. The removed lesion was submitted for histological examination.

On gross examination, the specimen consisted of multiple fragments of brownish tissue measuring 0.5–2 cm. All fragments were formalin fixed and paraffin embedded (FFPE) according to routine procedures.

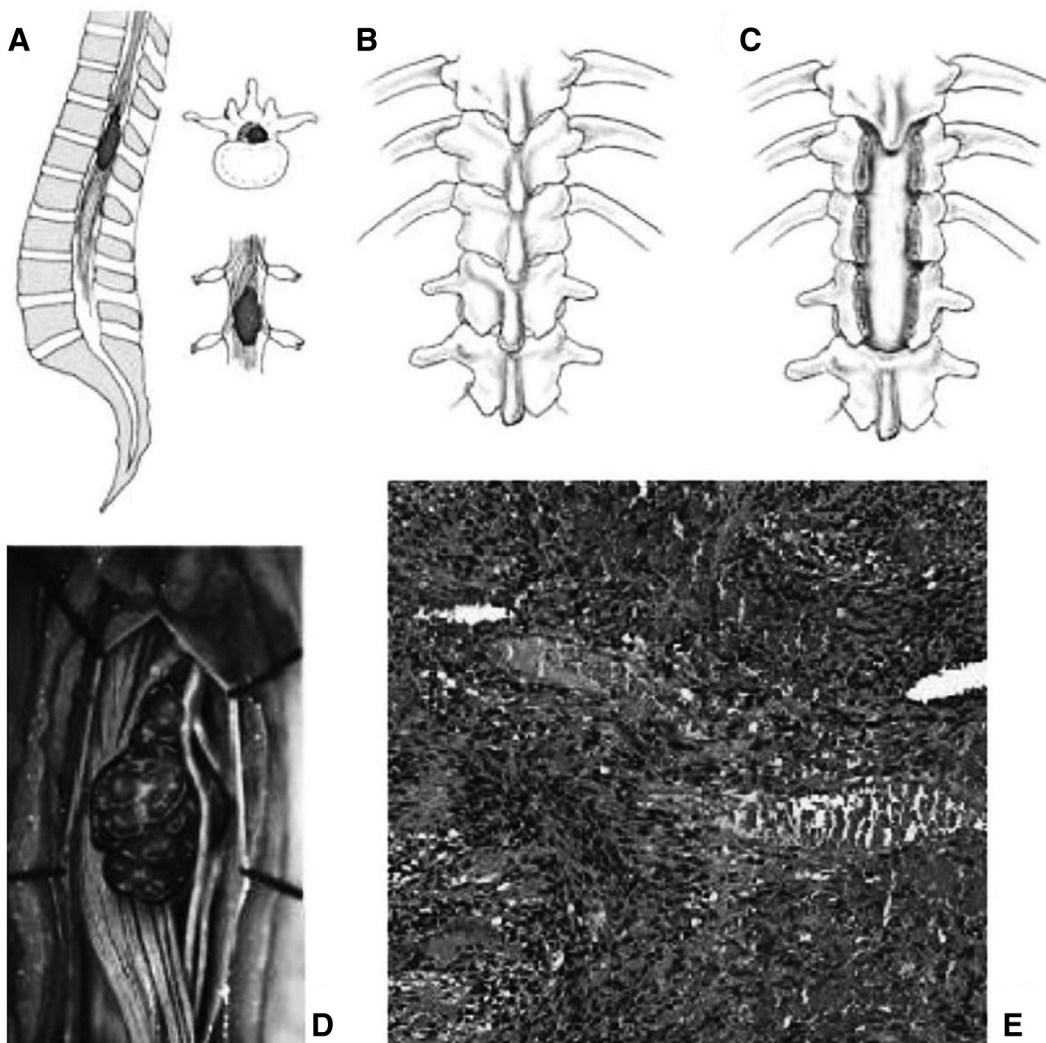


Fig. 2 Scheme of the lesion: Surgical approach: decompressive T11-L1 laminectomy and exposure of an extremely swollen dural sac, with ectatic vessels around (A–C). The mass presented intratumoral hemorrhage and displaced the roots of the cauda equina (D). Neoplastic spindle-cells, ectatic vessels, and diffuse intertumoral hemorrhage with hematoxylin-eosin (E).

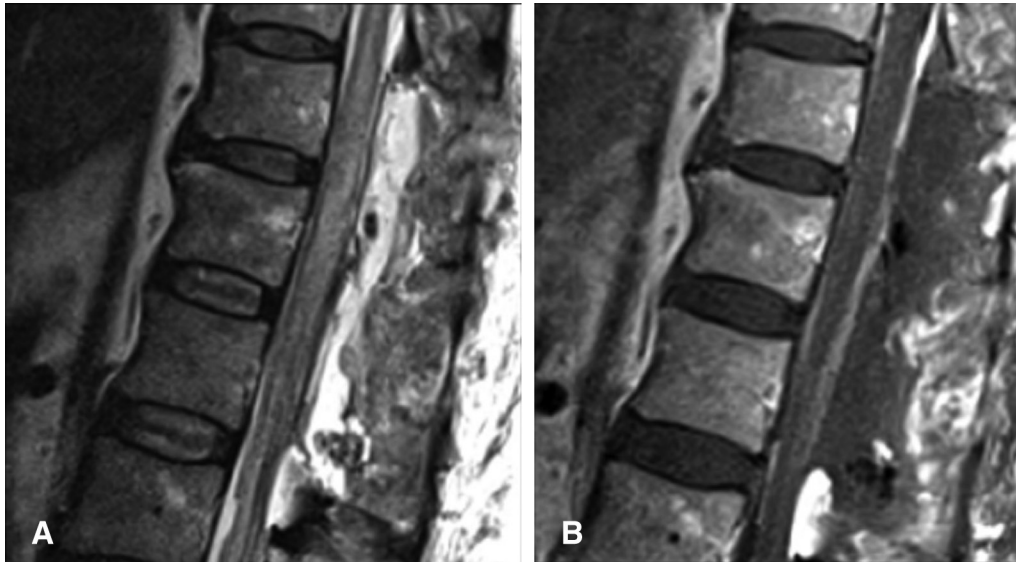


Fig. 3 Postoperative MRI. (A) Sagittal T2-weighted and (B) sagittal and T2-weighted (B) MRI showing the complete removal of the mass. The distal portion of the cord and conus medullaris, previously displaced, reveal mild swelling with T2 hyperintensity (A). MRI: magnetic resonance imaging.

On histology, the lesion was mostly composed of compact spindle cells organized in whorls and fascicles with sparse Verocay bodies. Some areas were loose-textured and showed myxoid appearance. Medium to large caliber ectatic vessels surrounded by extensive hemorrhage were present through the tumor (Fig. 2E). Small foci of coagulative necrosis were also noticed around degenerated vessels. The mitotic index was very low (<1 mitosis/50 high-power field [HPF]).

On immunohistochemistry, the neoplastic cells showed strong and diffuse S-100 protein positivity. Neurofilament immunostaining showed scattered neuronal axons mainly located at the periphery of the lesion. The proliferative index (evaluated on immunohistochemistry with anti-Ki-67 antibody) was 5%.

Therefore, the histologic diagnosis of hemorrhagic Schwannoma, grade I according to WHO 2016 Classification of Tumors of the Central Nervous System,¹⁾ was rendered.

After surgical treatment, the patient still exhibited a flaccid paraplegia (L1-L2 level) with sensory level around L3, while fecal and urinary incontinence persisted. Postoperative MRI showed that the lesion was completely removed, without surgical complications (Fig. 3). After 2 weeks from surgery, the patient declared a light improvement on tactile sensitivity of the front of the thighs. She was referred to a specialized center for intensive rehabilitation.

Discussion

Acute spinal subdural hemorrhage caused by spinal schwannomas is very rare: to the best of our

knowledge, 29 cases only have been previously reported in the English Literature (Table 1).^{2,4-30)} Most of our knowledge is based on single case reports: one series only of two cases has been reported.²²⁾

Based on Literature data, spinal hemorrhagic schwannomas affect more frequently adult patients, with a mean age at presentation of 51 years (ranging from 11 to 74). A slight female prevalence (M:F = 13:19) can be observed.

These hemorrhagic lesions affect almost equally the cervical spine (24%),^{4,5,8,11,18,25,27)} the lumbar tract (24%),^{7,13,16,17,21,22,28)} the thoracic tract (20%),^{2,6,9,14,19,29)} and the thoraco-lumbar passage (20%)^{10,12,20,22,24,26)} while the thoraco-cervical,¹⁵⁾ the sacral-lumbar tract,³⁰⁾ and the sacral spine²³⁾ are more rarely involved (12%). Bleeding can occur within the tumor (34%),^{6,11,12,17,22,25,26,27,28,30)} between the inner layer of the dura mater and the arachnoid mater of the meninges (subdural hematoma: 22%)^{4,5,8,9,16,18,19)} or into the subarachnoid space (subarachnoid hemorrhage: 16%)^{7,10,20,21,24)}; multiple sites can also be involved (13%), such as intratumoral bleeding with subdural hematoma^{2,23,29)} or subarachnoid hemorrhage.¹³⁾

The clinical picture is clearly different from that of spinal non-hemorrhagic schwannomas, commonly causing slowly progressive symptoms or spinal pain. Most frequent neurological symptoms at clinical presentation are acute and sudden (91%), due to the rapid compression of neural structures by bleeding from lesion; depending on the site involved, flaccid paraplegia, pain, urinary incontinence or retention, and impaired sensitivity depict the clinical presentation. Cases that showed

Table 1 Literature review of hemorrhagic spinal schwannoma

Author	Age/ sex	Clinical features	Duration of symptoms	Prior history	Spinal level	Enhancement on MRI	Type of hemorrhage
Smith, 1985 ⁴⁾	74/F	Cervical myelopathy	Acute	Absent	Cervical	Not performed	SDH
Vazquez- Barquero, 1994 ⁵⁾	68/M	Urinary retention, flaccid paraparesis, a decrease in right biceps and triceps jerks and a right Babinski's sign	3 days	Severe cervical and radicular pain along the upper limbs 3 days earlier	Cervical	Peripheral bright rim and central isointense signal	SDH
Uemura, 1998 ⁶⁾	58/F	Rapid progressive weakness	Sudden onset	Absent	T12	Peripheral	Intratumoral
Cordan, 1999 ⁷⁾	28/F	Progressive paraparesis and urinary retention	Subacute	Back pain for 10 days	L1-L2	N/A	SAH
Cohen, 2000 ²⁾	37/M	Flaccid paraplegia	24 hours	Minor fall	T11-12	Inhomogeneous	SDH Intratumoral
Ng, 2001 ⁸⁾	43/M	Upper limb pain and left hemiparesis	3 days	Absent	C6-7	Homogeneous	SDH
Tanaka, 2002 ⁹⁾	26/F	Severe lower limb weakness, and urinary retention	Sudden onset	Low backache for 3 years	T9-12	Absent	SDH
Parmar, 2004 ¹⁰⁾	56/M	Fever, neck pain, altered mental status, nuchal rigidity, mild symptoms of hesitancy, and poor stream of urine	Acute	Vague generalized backache for many years	Thora- columbar (T11-L1)	Heterogeneous enhancement	SAH
Ciappetta, 2008 ¹¹⁾	44/F	Neck pain, myelopathy, and upper limbs dysesthesia	3 days	Absent	FM-C5	N/A	Intratumoral
Ichinose, 2009 ¹²⁾	64/M	Backache, severe paraparesis, and urinary incontinence	Sudden onset	On anticoagulants	T12-L1	Peripheral	Intratumoral
Kukreja, 2014 ¹³⁾	47/M	Seizures and leg pain	Few days	Absent	L1-2	Heterogeneous	Intratumoral Intracranial SAH
Zhang, 2015 ¹⁴⁾	48/F	Severe leg pain and flaccid paraplegia	2 hours	Low backache for 1 year	T10-11	N/A	N/A
Prasad, 2016 ¹⁵⁾	40/M	Flaccid paraplegia	4 hours	Absent	C7-T3	Absent	N/A
Bennet, 2015 ¹⁶⁾	66/F	Lower extremity weakness and urinary incontinence	2 days	Chronic low back pain	L4	T1 hyperintensity, T2 hypointensity, without enhancement	SDH
Jenkins, 2015 ¹⁷⁾	62/M	Acute onset of severe pain and mild (4/5) weakness of right dorsiflexion	Sudden onset	Absent	L2-3	Minimal enhancement	Intratumoral

Table 1 Continued

Author	Age/ sex	Clinical features	Duration of symptoms	Prior history	Spinal level	Enhancement on MRI	Type of hemorrhage
Sahoo, 2015 ¹⁸⁾	44/M	Acute quadripareisis	Sudden onset	Absent	C3-C5	Heterogeneous	SDH
Hdeib, 2016 ¹⁹⁾	71/M	Severe lower limbs weakness and urinary retention	Sudden onset	Back pain	T8	N/A	SDH
Zhang, 2016 ²⁰⁾	51/M	Paraplegia	Subacute	Back pain for the last 3 days	T11-L1	Not performed	SAH
Zhang, 2016 ²¹⁾	47/F	Symptoms mimicking meningitis	Subacute	Fever and headache for 2 weeks	T9	Homogeneous on T1-weighted images, heterogeneous enhancement on T2	SAH
Kimura, 2018 ²²⁾	64/F 61/F	Low back pain Low back pain and numbness of both leg	Subacute Chronic	Low back pain for 10 days after falling on her buttocks Low back pain for 6 years. Antiplatelet therapy for acute coronary syndrome	T12-L1 L2-L3	Isointense to hypointense on T1-weighted images Hypointense on T1-weighted images, heterogeneous enhancement on T2	Intratumoral Intratumoral
Naadem, 2017 ²³⁾	68/F	After trauma: back pain, lower extremity weakness, and urinary incontinence	Sudden onset	N/A	Conus medullaris	Heterogeneous	SDH Intratumoral
Tanki, 2018 ²⁴⁾	11/F	Symptoms mimicking meningitis	Acute	N/A	T12-L1	Isointense to hypointense on T1-weighted images, mildly enhancing	SAH
Gandhoke, 2018 ²⁵⁾	38/M	Spastic quadripareisis	Acute	Neck pain radiating to the left upper extremity for the last 8 months	C2-C4	Hypointense on T2, a heterogeneously hyperintense solid-cystic	Intratumoral
Rahyussalim, 2019 ²⁶⁾	38/F	Lower limbs weakness, impaired sensitivity, defecating, and urinating problems	2 months after the beginning of treatment with only joint manipulation	Difficulty standing up from squatting positions; heaviness, and numbness from hips radiated to knees and ankles for 2 years	T10-T12, grown to T10-L2	Hyperintensity	Intratumoral
Jung, 2019 ²⁷⁾	37/M	Neck pain, quadripareisis more severe in the right-side limbs, decreased pain sensation in the left side limbs, and decreased touch sensation in the right-side limbs	Acute onset after physical therapy	Prior clinical suspicion of cervical myelopathy	C2-C3, extending to the right side of the cord	Heterogeneously mixed signal intensity mass and mild enhancement	Intratumoral

Table 1 Continued

Author	Age/ sex	Clinical features	Duration of symptoms	Prior history	Spinal level	Enhancement on MRI	Type of hemorrhage
Gotecha, 2019 ²⁸⁾	61/F	Chronic low backache	3 years after radicular symptoms	Previous radicular symptoms	L3-L5	Hyperintensity	Intratumoral
Dobran, 2019 ²⁹⁾	38/M	Lower limb plegia, and urinary retention	Acute after minor trauma	N/A	T11	Heterogeneous	Intratumoral, SDH
Ito, 2019 ³⁰⁾	58/F	Pain in her back and her left leg	Acute	N/A	L4/L5/S1	Heterogeneous	Intratumoral
Current case	57/F	Flaccid paraplegia, impaired sensitivity, and fecal and urinary incontinence	Less than 24 hours after the beginning of severe lumbar pain and right limb weakness	Hypertension	Thora- columbar (T11-L1), arising from Cauda Equina	Heterogeneous enhancement	SDH Intratumoral

N/A: not available, SAH: subarachnoid hemorrhage, SDH: subdural hemorrhage.

subarachnoid hemorrhage are likely to present findings mimicking meningitis.^{10,21,24)} Therefore, cerebrospinal fluid examination can be of help to differentiate meningitis from hemorrhage, in cases presenting a spinal cord mass with meningeal irritation signs (headache, fever, neck stiffness, and positive Kerning sign).

The pathogenesis of acute hemorrhage in spinal schwannomas and other spinal cord tumor, such as neurofibroma³¹⁾ or myxopapillary ependymoma,³²⁾ is not fully understood. Two possible theories have been suggested: the vascular theory and the mechanical one. The first one supports the hypothesis that intratumoral abnormal dilated vessels may undergo a spontaneous thrombosis or rupture, resulting in bleeding.^{33,34)} The second theory states that movements of the spine may induce traction on tumor vessels, causing their sudden rupture.^{8,32)} Spinal traumas may aggravate these situations while an adjunctive risk-factor may be an anticoagulant/antiplatelet therapy.²⁷⁾

Considering the histological features of our specimen (intratumoral dilated blood vessels showing tortuous course, with fragile, hyalinized walls, and focal ischemic damage), the clinical history of the patient (no history of trauma), and the intraoperative appearance of the lesion (attached to an ectatic but intact vessel perforating the dura mater), we hypothesize that extensive bleeding might be associated with spontaneous rupture of intratumoral blood vessels. Bleeding started within the lesion at first and then involved the subdural space.

Preoperative diagnosis can be very difficult because clinical and radiological features of an hemorrhagic schwannoma are non-specific and can mimic the diagnostic picture of several lesions.

In this case, the MRI of the dorsal and lumbar spine suggested an hemorrhagic intradural extramedullary lesion.

The differential diagnosis may include vascular malformations (cavernous hemangiomas, arteriovenous fistulas), benign intramedullary tumors (ependymoma, hemangioblastoma, paraganglioma, glomic tumors), benign extramedullary neoplasms (meningiomas), and malignant tumors (metastasis). Histological examination together with a specific immunohistochemical stain panel is required for definite diagnosis and it should include epithelial (wide spectrum cytokeratins), glial (glial fibrillary acidic protein [GFAP], Olig2), meningeal (epithelial membrane antigen [EMA], Progesterone Receptor), vascular (CD34), neuroendocrine (Synaptophysin), and melanoma markers.

In conclusion, the diagnosis of a spinal hemorrhagic schwannoma, although uncommon, should be always kept in mind in case of a patient presenting with acute neurological deterioration due to a spinal intradural extramedullary bleeding involving the neural structures or the cauda equina. The definitive diagnosis is obtained only after the surgical excision of the lesion and its histological examination.

Conflicts of Interest Disclosure

The authors declare no conflict of interest regarding the present paper.

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