Lumbar Spinal Epidural Capillary Hemangioma: A Case Report and Literature Review

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Patient: Female, 38-year-old
Final Diagnosis: Spinal capillary hemangioma
Symptoms: Leg cramp • low back pain • paresthesia • unsteady gait
Medication: —
Clinical Procedure: Surgical resection
Specialty: Radiology

Objective: Rare disease
Background: Capillary hemangiomas are often seen on the skin of young individuals and are rarely found in the spine. These vascular lesions can arise from any spinal compartment, although they are more commonly found in the intradural extramedullary (IDEM) than the epidural location. We present a unique case of a woman with a histologically proven spinal epidural capillary hemangioma (SECH). The imaging and histopathological characteristics, as well as the treatment strategy of this vascular lesion, are highlighted along with a comprehensive review of the literature.

Case Report: A 38-year-old woman presented with progressively worsening low back pain that radiated to both legs. Neurological examination revealed a weakness of the left leg without sensory loss. Magnetic resonance imaging (MRI) demonstrated an epidural tumor at L1-L2 level, making an obtuse angle with the cerebrospinal fluid (CSF) on sagittal T2-weighted images. The patient underwent a complete tumor resection without complications or recurrence. The histology revealed a capillary hemangioma.

Conclusions: SECH is exceedingly rare, with only 22 cases in the reported literature. Females are more commonly affected than males, and the thoracic spine is more commonly involved than the lumbar spine. SECH often mimics other epidural and IDEM lesions, leading to misdiagnosis. MRI is useful to differentiate SECH from lesions in the various spinal compartments; additionally, MRI is essential for preoperative planning and patient surveillance. Preoperative embolization is an option given the high vascularity of SECH. Surgery is the mainstay treatment, with a good prognosis, in most cases without recurrence.

Keywords: Epidural Neoplasms • Hemangioblastoma • Hemangioma, Capillary • Paraganglioma

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Background

Hemangiomas are congenital vascular malformations, classified according to the predominant vascular morphology: cavernous, capillary, arteriovenous or venous [1]. Capillary hemangiomas are the most common subtype [1]. They occur frequently in the skin and soft tissues of younger individuals and are rarely found in the central nervous system (CNS) [2,3]. Capillary hemangiomas can arise from any compartment of the spine, although they are more commonly found in the intradural extramedullary (IDEM) location [2]. Spinal epidural capillary hemangiomas (SECH) are exceedingly rare. We here present a unique case of a woman with a histologically proven SECH in the upper lumbar spine. The imaging and histopathological characteristics, as well as treatment strategy of this abnormally located vascular lesion in the spine, are highlighted along with a comprehensive review of the literature.

Case Report

A 38-year-old ethnic Malay woman presented to the emergency department with progressive worsening low back pain and radiating pain to both thighs and the lateral aspect of the left calf. In addition, she was concerned about her unsteady gait and episodes of leg cramps followed by ‘pin and needles’ paraesthesia in both thighs, including the knees. Her symptoms had worsened over the last 3 months such that she had difficulty riding a motorcycle. She had no significant past medical history. Neurological examination revealed a diffuse mild weakness of the muscles of the left leg (4/5) without any loss of sensation to pain or temperature. The initial lumbar spine radiograph was unremarkable. She was subsequently seen at the outpatient orthopaedic service with persistent symptoms.

Magnetic resonance imaging (MRI) of the lumbar spine (Figure 1A-1G) was performed using a 1.5 Tesla MR system (MAGNETOM Avanto, Siemens, Erlangen, Germany). The following MRI protocol was conducted: axial and sagittal T1- and T2-weighted, and post gadolinium axial and sagittal T1-weighted fat-saturated sequences. MRI showed a well-defined and homogeneous enhancing lesion in the posterior aspect of the spinal canal at the L1-L2 level, measuring approximately 1.1×2.0×5.0 cm (anteroposterior x transverse x craniocaudal) in size. The lesion formed an obtuse angle with the adjacent cerebrospinal fluid (CSF) on the sagittal T2-weighted image (Figure 1B). There were no significant intratumoral flow voids. Sagittal (Figure 1B) and axial T2w (Figure 1E) images showed
anterior displacement and compression of the cauda equina and the posterior dura layer by the tumor. As the epidural lesion was covered by an overlying dura, it has the appearance of a “marble under the carpet” where the marble represents the tumor (arrow) and the carpet resembles the dura mater [4].

Surgical resection was indicated based on the progression of the patient’s symptoms and the degrees of compression on the cauda equina. The patient underwent L1-L2 laminectomy with complete tumor excision without complications. Preoperative embolization was not deemed necessary given the lack of intratumoral flow voids on the diagnostic MRI study. Histopathological examination revealed a capillary hemangioma (Figure 2A, 2B). Oral analgesics and intravenous opioids were administered for postoperative pain management. There was a gradual improvement of her gait and reduction of paresthesia in her legs. She was discharged on the fourth postoperative day after a short course of physiotherapy and

Figure 1. Magnetic resonance imaging (MRI) of a 38-year-old woman with spinal capillary hemangioma. (A) Sagittal T1-weighted (w) non-contrast image shows a well-defined T1 iso- and hyperintensity lesion relative to the spinal cord at the L1-L2 level (arrow). The tumor effaces the posterior epidural fat (arrowheads). (B) Sagittal T2w image shows a hyperintense extradural tumor (arrow) forming an obtuse angle (red-dotted lines and elbow connector arrow) with adjacent CSF. As the extradural tumor is covered anteriorly by an overlying dura, it gives the appearance of “marble under the carpet” where the marble represents the tumor (arrow) and the carpet resembles the dura mater. (C) Sagittal T1w fat-saturated post-contrast image reveals a well-defined and avid enhancing extradural tumor (arrow) displacing cauda equina nerve roots anteriorly within the spinal canal. The tumor effaces the posterior epidural fat without involving the posterior elements of the vertebrae (arrowheads). (D) Axial T1w pre-contrast image shows the extradural tumor (arrow) symmetrically displacing and indenting dura and cauda equina anteriorly. (E, F) Axial T2w images demonstrate the T2 hyperintense tumor (arrows) causing anterior displacement of the cauda equina nerve roots (arrowhead) within the spinal canal. The intraspinal nerve roots are arranged in a curvilinear fashion, surrounded by a thin rim of CSF at L1-2 level (arrowheads). (G) Axial T1w fat-saturated and post-contrast image shows the well-defined and avid enhancing extradural lesion (arrow) measuring approximately 1.1×2.0×5.0 cm (anteroposterior x transverse x craniocaudal) in size. It displaces the cauda equina nerve roots anteriorly within the spinal canal. The intraspinal nerve roots are arranged in a curvilinear fashion, surrounded by a thin rim of CSF at L1-2 level (arrowhead).

Figure 2. (A) Lower magnification photomicrograph showing numerous small and thin-walled capillaries lined by single-layered bland endothelia separated by mature adipose tissue. There is no evidence of cytologic atypia [Hematoxylin and eosin (H&E) stain magnification, ×10]. Histological findings are compatible with a capillary hemangioma with fatty involution. (B) Higher magnification photomicrograph showing again numerous thin-walled capillaries lined by single-layered bland endothelia with fatty involution. No evidence of cytologic atypia is detected (H&E stain, magnification, ×20). Histological findings are compatible with a capillary hemangioma with fatty involution.
occupational therapy. There was no evidence of clinical or radiological recurrence over 3 years of follow-up after surgery.

Discussion

Capillary hemangiomas in the epidural location are exceedingly rare, with only 22 reported cases (including our current case) thus far in the literature (Table 1). Most patients present in their fourth to fifth decades of life, with ages ranging from 17 to 69 years and a median age of 49.2 years (Table 1). There is a slight predilection for females, with the male-to-female ratio of 9:13. These benign vascular lesions occur more frequently in the thoracic (16/22, 72%) than the lumbar spine (6/22, 27%) (Table 1). Half of the cases showed foraminal extension, which allowed differentiation from cavernous hemangioma [1]. Owing to the slow-growing nature of SECH, most patients present with back pain, progressive myelopathy, or radiculopathy (Table 1) [2,5]. There were a few post-traumatic-related cases of SECH without a definite pathophysiological cause identified.

Capillary hemangiomas can arise from the vasa nervosum or blood vessels of the nerve roots in the cauda equina, the surface of the dura, or the pial surface of the spinal cord, but none were found within the spinal cord [2]. Histologically, capillary hemangiomas are characterized by lobules of thin-walled and irregular capillary-sized vessels lined by endothelial cells, enveloped by fibrous stroma or capsule. They are often associated with feeding vessels [6].

MRI is the primary imaging modality for diagnosis, pre-treatment planning, and surveillance of all spinal lesions, including SECH (Figure 3). The advantages of MRI are the ability to narrow the differential diagnosis by defining the tumor location, signal characteristics, and the relationship of the mass to the cord, dura, and nerve root [7]. In addition, it can help to identify multiple or secondary lesions along the spine, including metastatic deposits, drop metastases, and large feeding or draining vessels [8]. Typical MR imaging features of capillary hemangioma include well-circumscribed tumor margins and typical location in the posterior portion of the spinal canal. SECH can occasionally efface the posterior epidural fat of the spine without involving the posterior elements of the vertebra (Figure 1C). The tumor often demonstrates T1-weighted isointensity relative to the spinal cord and T2-weighted hyperintensity, as well as avid and homogeneous post-contrast enhancement [2,8,9]. The presence of flow voids and/or avid enhancement of the tumor is reflective of its vascular nature [10]. These features are, however, non-specific, hence it can be challenging to distinguish SECH from other epidural tumors on imaging. Histologic confirmation is necessary in most cases. Examples of other spinal epidural tumors include metastases, multiple myeloma, lymphoma, hemangiopericytoma, meningioma, Ewing’s sarcoma, and myeloid sarcoma (Table 2) [11]. Common metastases to the spine usually originate from a non-CNS primary malignancy (eg, breast, lung, prostate, renal, gastrointestinal and thyroid carcinoma) [12]. The common non-neoplastic lesions found in the epidural compartment include disc herniation, epidural hematoma, lipomatosis, venous plexus, and abscess (Figure 3) [13].

Capillary hemangiomas are also found in the intradural extramedullary (IDEM) compartment with a predilection for the conus and the lumbar spine [2]. It can be challenging to identify the correct compartment from where the lesion arises. Epidural tumors commonly obliterate the CSF space within the spinal canal, unlike IDEM, in which the surrounding CSF space is preserved (Table 2) [2]. Additionally, the dura and the thecal sac would be displaced away, from the epidural mass. An obtuse angle is typically formed between the epidural mass and the adjacent CSF with a ‘marble under the carpet’ appearance on sagittal T2w images; the marble represents the tumor and the carpet resembles the overlying dura mater [4]. The cord may appear widened in one plane due to pressure from the epidural mass, surrounded by a thin layer of contrast [4].

Conversely, IDEM tumors commonly form an acute angle with the cord/CSF on sagittal T2w images, giving the appearance of “marble on the carpet” (Figures 1, 3).

Capillary hemangiomas are often misdiagnosed radiologically as being meningiomas and schwannomas; all of them demonstrate avid post-contrast enhancement [9]. Meningioma usually shows T1-weighted iso- or slight hypointensity, and iso- or slight T2 hyperintensity [9]. The ‘dural-tail sign’ is not always a reliable feature to differentiate between meningioma and capillary hemangioma, since the latter occasionally arises from the surface of the dura mater, resembling a dura-tail. Other imaging characteristics of meningioma such as the ‘CSF cleft sign’ and bony hyperostosis are perhaps more useful distinguishing features. Schwannomas are typically hypointense on T1-weighted and hyperintense on T2-weighted images with occasional cystic change or necrosis, which can distinguish them from capillary hemangioma [9]. The precise preoperative differentiation between meningioma/schwannoma from capillary hemangiomas is not crucial to management, since these are non-aggressive tumors. Vascular lesions in the spine with flow voids often mimic capillary hemangioma and vice versa; these include paraganglioma, intradural arteriovenous malformation/fistula (AVM/AVF), and hemangioendothelioma [10]. Paragangliomas contain vascular flow voids in the form of ‘salt-and-pepper’ appearance (unlike capillary hemangiomas), while hemangioendothelioma involves the vertebral bodies as osteolytic lesions with soft tissue components.
| First Author, year | Age (years)/gender | Presenting symptoms | Epidural location | Other (MRI) features of tumor | Laminectomy, resection | Outcomes |
|-------------------|-------------------|---------------------|------------------|-----------------------------|----------------------|----------|
| Gupta, 1996 [17]  | 50/M              | BP, myelopathy, weakness LL | T8-10            | Post-contrast enhancement   | TR               | NR, GR   |
| Badinand, 2003 [18]| 40/F              | Gait disturbance, LL pain, incontinence | T2-4            | T3-4 foraminal extension | TR               | NR, GR   |
| Kang, 2006 [19]   | 56/M              | Radiating chest wall pain | T2-4            | Dumbbell-shaped             | PR               | NR, GR   |
| Tekin, 2008 [20]  | 56/F              | BP, radiates to R thigh, L4 hypoesthesia | L3-4            | Heterogeneous with flow voids | TR               | NR, symptoms improved |
| Akhaddar, 2010 [21]| 19/F              | BP, progr gait disturbance | T5-6            | Homogeneously-enhanced, foraminal extension | TR               | NR, GR   |
| Hasan, 2011 [22]  | 57/M              | BP, progr myelopathy, weakness LL | T10-12          | Foraminal extension         | PR               | NR, symptoms improved |
| Shilton, 2011 [23]| 47/F              | BP, thoracic myelopathy (post-traumatic) | T7-8            | Foraminal extension         | TR               | NR, symptoms improved |
| Vassal, 2011 [24] | 59/F              | BP, R intercostal neuralgia | T5-7            | Intrathoracic extension     | TR               | NR, GR   |
| Seferi, 2014 [25] | 58/M              | BP, paraparesis, sensory disturbance | T2-4            | Foraminal extension         | TR               | NR, GR   |
| Gencpinar, 2014 [26]| 17/F             | Weakness LL               | T3-7            | Foraminal extension         | TR               | NR, GR   |
| Garcia-Pallero, 2015 [27]| 67/F   | Dyspnea, pleural effusion | T4-5            | Heterogeneous paravertebral tumor with intrathoracic extension | TR               | NR, GR   |
| Egu, 2016 [28]    | 60/F              | BP, S1 radiculopathy     | L5-51           | Homogeneous enhancing       | TR               | NR, symptoms improved |
| Kilic, 2017 [29]  | 40/M              | BP, L LL pain (post-traumatic) | L1              | Vertebral body, pedicle and transverse process | TR               | NR       |
| Rajeev, 2017 [30] | 50/M              | Paraparesis LL            | L1-2            | Enhancing mass, foraminal extension | TR               | NR, symptoms improved |
| Brasil, 2018 [1]  | 69/F              | BP                          | T9-10           | No extension                | TR               | NR       |
| Xu, 2018 [12]     | 57/M              | BP, LL weakness            | T2-3            | Foraminal extension         | TR               | NR, symptoms improved |
| Cofano, 2019 [31] | 52/F              | BP                          | T6-9            | Foraminal extension         | TR               | NR       |
| Sudir, 2019 [32]  | 63/M              | BP, LL weakness (post-traumatic) | T6-8            | Vertebral body, pedicle and transverse process | TR               | NR       |
Table 1 continued. Summary of all the reported cases of spinal epidural capillary hemangioma in the literature.

| First Author, year | Age (years)/gender | Presenting symptoms | Epidural location | Other (MRI) features of tumor | Lamincetomy, resection | Outcomes |
|-------------------|--------------------|---------------------|------------------|-----------------------------|------------------------|----------|
| Niznick, 2020 [14] | 51/F               | Sensory and gait disturbances | T5-6             | Foraminal extension          | TR                     | NR       |
| Rajpal, 2020 [15]  | 29/F               | Flank pain          | T7               | Foraminal and intrathoracic extension | TR                     | NR       |
| Doi, 2021 [33]     | 47/M               | BP, LL pain, sensory disturbance, incontinence | L2-4             | Also involves spinous process and laminae | PR                     | NR       |
| Current case       | 38/F               | Progr BP, gait disturbance, LL weakness and paresthesia | L1-2             | Homogeneous enhancement      | TR                     | NR, symptoms improved |

BP – backpain; GR – good recovery; F – female; L – left; LL – lower leg, M – male; NR – no recurrence or residual tumor; progr – progressive; PR – partial resection; R – right; TR – total resection.

Figure 3. Schematic diagram of the spinal MRI (created using Microsoft PowerPoint drawing function) illustrated the various spinal lesions distributed in different compartments within the spine. Intramedullary lesions are often associated with spinal cord expansion, while extramedullary lesions narrow the calibre of the spinal cord. Extramedullary lesions are further divided into those commonly found in the intradural extramedullary and extradural compartments by virtue of the state of the surrounding CSF and the angle between the lesions and cord/CSF space. Intradural extramedullary lesion compresses the spinal cord and forms an acute angle with the cord and gives the appearance of a “marble above the carpet”; the marble represents the tumor/lesion and the carpet resembles the dura mater. Besides, the CSF space is relatively preserved and there is a smooth interface between the cord and the tumor. Extradural lesions often efface the surrounding CSF space and form an obtuse angle with the CSF on sagittal T2w image. It gives the appearance of a “marble under the carpet”. AVM – arteriovenous malformation; AVF – arteriovenous fistula; CSF – cerebrospinal fluid; mets – metastases.
### Table 2. Differentiating spinal epidural capillary hemangioma from other epidural and intradural extramedullary tumors.

| Compartment | Spinal tumors                        | Prevalent age-group | Typical location | Pre-contrast MR signal intensity | Post-contrast enhancement | Other imaging features |
|-------------|--------------------------------------|---------------------|------------------|---------------------------------|--------------------------|------------------------|
| Epidural    | Metastases                           | Incidence ↑ with age (typically >40 years old) | Predilection for the posterior elements. | T1↓ T2↑↔ | Enhances | Predilection for the posterior elements, may have cortical destruction with associated soft tissue mass. Commonly seen in breast, lung, prostate, renal, gastrointestinal, and thyroid |
| Multiple myeloma (MM) | E. Ads M>F | Any vertebral body, typically sparing the pedicles (vs metastases/malignancy-pedicles involved) [16] | Osseous (T2↑ T1↓) Extraosseous T2↓ | Avid Homogeneous | Extraosseous lesions contiguous with bone, often larger and occur in a paraspinal or epidural location. MM can be associated with collapsed vertebral body, and cord compression |
| Ewing sarcoma | Chd, Y. Ads | Thoraco-Lumbar | T2↑ T1↓ | Heterogeneous enhancement of tumor and affected vertebrae | CT scan: Lytic permeative destruction |
| Primary vertebral lymphoma (commonly NHL type) | Ads (5th-7th decades), M>F (8: 1) | Lesions arise from epidural lymphoid tissue along the thoracic and lumbar spine | T2↑ | Homogeneous | Restricted diffusion with markedly low ADC |
| Spinal epidural capillary hemangioma | Ads (4th-5th decades) M>F (9: 13) | Thoracic > lumbar spine (16: 6) | T2↑ T1↓ | Homogeneous | Foraminal extension in 50% of cases |
| SFT/HPC | Ads (4-6 decades) M>F | Thoracic > cervical > lumbar | T1++, T1↓ T2↑ | Intense homogeneous |
| IDEM | Meningioma | Ads, F>M (9: 1) | Thoracic > cervical | T2++, T2↑ | Intense homogeneous | Broad-based dural attachment and CSF cleft sign. Commonly – solitary; multiple lesions in MEN type II |
| Schwannoma | Y. Ads M>F | Dorsal nerve root, neural foramen | Thoracic > cervical | Homogeneous or inhomogeneous | Cysts and necrosis. Commonly – solitary; multiple lesions in MEN type II |
Table 2 continued. Differentiating spinal epidural capillary hemangioma from other epidural and intradural extramedullary tumors.

| Compartment                        | Spinal tumors | Prevalent age-group | Typical location       | Pre-contrast MR signal intensity | Post-contrast enhancement | Other imaging features                                      |
|------------------------------------|---------------|---------------------|------------------------|---------------------------------|--------------------------|------------------------------------------------------------|
| Paraganglioma                      | Ads M>F       | Conus, cauda equina | T2↔, T2↑              | Mildly inhomogeneous “salt-and-pepper” appearance due to prominent flow voids | Hemosiderin from prior hemorrhage (T2↓ rim “cap sign”) detected on T2 and GRE sequences |
| Myxopapillary ependymoma (13% of all spinal ependymomas) | Y. Ads, M>F   | Conus medullaris, cauda equina | T2↑ at tumor margin (hemosiderin) | Typically homogeneous (except hemorrhage) | Hemorrhage detected on GRE sequences |

ADC – apparent diffusion coefficient; Adol – adolescent; Ads – adults; Chd – children; CT – computed tomography; E – elderly; F – females; GRE – T2*-weighted gradient-recorded echo; HPC – hemangiopericytoma; IDEM – intradural extramedullary; M – men; MEN – multiple endocrine neoplasia; MR – magnetic resonance; NHL – non-Hodgkin’s lymphoma; SFT – solitary fibrous tumor; Y – young; symbols: ↑ – hyperintense; ↓ – hypointense; ↔ – isointense.

in the epidural space [10]. AVM/AVF typically appear as serpiginous structures in the perimedullary regions of the spine (Table 2) [10].

Analysis of the configuration and location of tumors on MRI is highly important for surgical planning and to guide tumor resection (Table 2). In addition, the tumor extension and vascularity can be better assessed on MRI [3,8].

Although considered benign, capillary hemangiomas can cause spinal cord or cauda equina compression, as well as foraminal extension ([15] – see Table 1). There is also the risk of rare hemorrhagic complications due to the vascular nature of the lesion. Surgery should be considered even in the absence of neurological symptoms. To reduce intraoperative hemorrhage, preoperative angiography and/or embolization can be considered [3,6]. In our case, preoperative embolization was not deemed necessary given the lack of intralesional flow voids on the diagnostic MRI study. With adequate preoperative planning, total surgical resection can be achieved in most cases by carefully dissecting the lesion away from the dura while exercising judicious hemostasis by eliminating all arterial feeders. The prognosis is good, in most cases with no recurrence (Table 1).

Conclusions

MRI is essential for diagnosis, preoperative planning, and patient surveillance. Knowledge of the MRI characteristics of spinal lesions and their locations within the spinal compartments can help narrow their differential diagnosis. SECH can be differentiated from IDEM lesions, whereby these lesions form obtuse and acute angles with the CSF, respectively. Some SECH demonstrate foraminal extension distinguishing them from cavernous hemangioma. Nevertheless, surgical resection is the mainstay treatment for capillary hemangioma, with a good prognosis, in most cases without recurrence.

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Department and Institution Where Work Was Done

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Declaration of Figures’ Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.
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