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Case Report

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Rapid growth of spinal epidural capillary hemangioma associated with isolated intraosseous lesion at the same level: A case report

Abstract

A 47-year-old man presented with a spinal epidural capillary hemangioma manifesting as a 6-month history of worsening low back pain and lower limb pain. Computed tomography and magnetic resonance imaging (MRI) at a previous hospital showed intraosseous abnormal signals and an epidural lesion at the L3 level. One month later, MRI revealed rapid growth of the epidural lesion. Total resection of the epidural tumor and partial removal of the intraosseous tumor were performed. Macroscopically, the intraosseous lesion and epidural lesion seemed to be isolated from each other. Histopathological diagnosis was consistent with capillary hemangioma. Spinal epidural capillary hemangioma sometimes recurs via underlying arteriovenous anastomosis. Progressive clinical symptoms, as seen in our case, indicate the possibility of tumor growth even if the lesions are well demarcated, which suggests of benign tumor. Spinal epidural capillary hemangioma in addition to metastatic spinal tumors should be considered in the differential diagnosis of spinal epidural tumors associated with intraosseous lesions.

Keywords: capillary hemangioma, rapid growth, epidural, arteriovenous anastomosis, bony involvement

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Introduction

Most spinal epidural tumors are nerve sheath tumors (NSTs) and metastatic spinal tumors, some of which are hemangiomas. Most spinal epidural hemangiomas are cavernous hemangiomas, although 20 cases of spinal epidural capillary hemangioma (SECH) have been reported.[1–20] Most SECH cases had foraminal extension, which easily allowed differentiation from cavernous hemangioma.[9] SECH cases tended to occur in the thoracic levels and were rare in the lumbosacral regions. Only two of the 20 cases of SECH were associated with intraosseous lesions.[11,17] However, these epidural lesions were thought to result from adjacent bone destruction after spinal trauma. We report an extremely rare case of SECH associated with isolated intraosseous lesion at the same level despite the absence of inherited disorders, malignant tumors, and spinal trauma. The tumor underwent rapid growth in a short period. We report the features of the SECH and the probable mechanisms of rapid growth.

Case description

A 47-year-old male presented with a 6-month history of worsening low back and lower limb pain. He had no family history or previous history of malignant tumor or spinal trauma. Previous examination at another hospital found urinary incontinence and sensory disturbance from the lower limbs to anus associated with the pain. Computed tomography showed intraosseous abnormal signals in the bilateral laminae and spinal process at the L3 level (Figure 1A-1C). Magnetic resonance imaging (MRI) also revealed an epidural lesion with a maximum size of 23 mm at the same level (Figure 1D-1F). On admission to our hospital one month later, he showed similar urinary incontinence without motor deficits, but sensory disturbance had extended to the posterior surface of the thighs with pain. MRI demonstrated no changes in the intraosseous lesion but rapid growth of the epidural lesion with a maximum size of 74 mm extending over the L2-L4 levels within only one month (Figure 2A-2C). The epidural lesion seemed to be radiologically isolated from the intraosseous lesion. T1-weighted MRI showed the epidural lesion as mixed-intensity areas inside capsular barriers. T2-weighted imaging and short T1 inversion recovery imaging also showed the lesion as hyperintense (Figure 2A-2C, 2E). The lesion was almost homogeneously enhanced (Figure 2D). Preoperative diagnoses were NST or metastatic spinal tumor of unknown origin.

We planned tumor resection via the posterior approach, based on the radiologically rapid growth of the lesion and progression of his clinical symptoms. The patient was placed in the prone position under general anesthesia with motor evoked potential monitoring. Posterior midline skin incision above the L3 spinal process was performed. Removal of the spinal process at the L3 level exposed the soft lesion in the apparently loose trabecular bone with bleeding (Figure 3A). After removing the L3 laminae, the reddish highly vascularized lesion was found to be covered with epidural fat tissue and a thin capsule (Figure 3B). This epidural lesion was well-demarcated without attachments. The lesion could be easily detached from the adjacent tissue and dura mater (Figure 3C). Macroscopically, the intraosseous lesion did not extend outside the cortical bone. The radiological lesions were resected as far as possible because they were not invaded by the epidural tumor. Total resection of the epidural tumor and partial removal of the intraosseous tumor were performed. Partial laminectomies ranging from the lower L2 to upper L4 levels were also performed.

The lesion consisted of blood vessels and mesenchymal spindle-shaped cells intermingled with the vessels. These vessels had thin-walled lobular architecture with variable caliber. The vessels were poorly circumscribed and arranged irregularly (Figure 4A). No evidence of macrophages suggestive of inflammation was found. Immunohistological staining showed the endothelial cells were positive for CD31, CD34, and vimentin, but negative for epithelial membrane antigen and somatostatin receptor 2A. The MIB-1 index

was approximately 2-3% (Figure 4B). These histopathological findings were consistent with SECH.

Postoperatively, he had an eventful course. His low back pain and urinary incontinence gradually improved, but numbness remained. MRI at 2 months postoperatively showed that the epidural lesion had completely disappeared, but abnormal intraosseous signals in the bilateral pedicles were seen, suggestive of tumor remnant (Figure 4C). He has continued follow up as an outpatient.

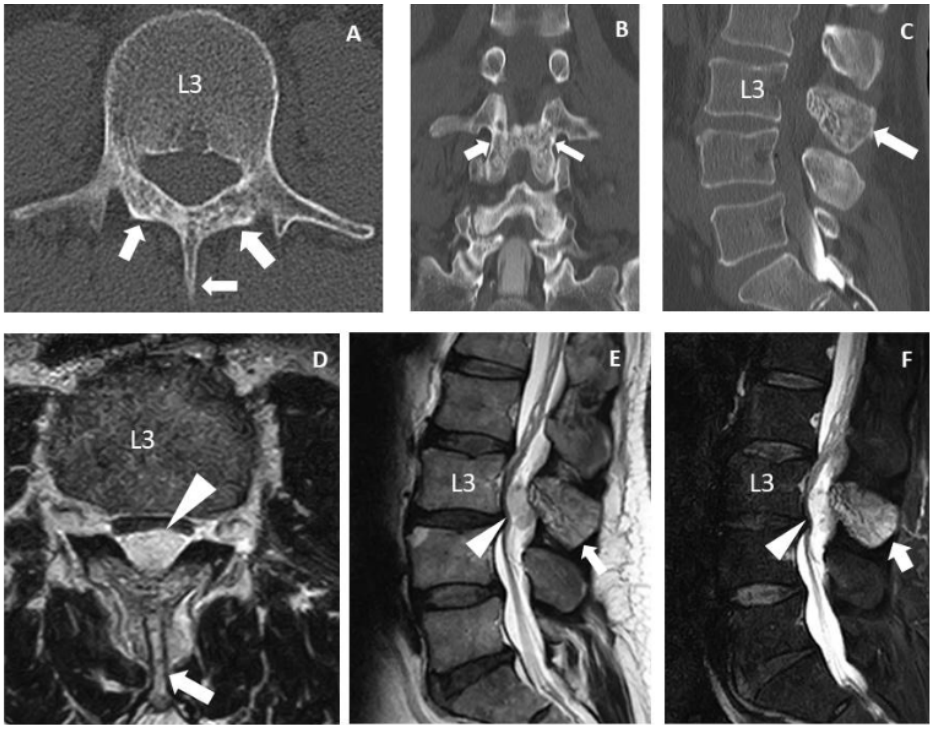

Figure 1 Images obtained at the previous hospital. Computed tomography (CT) scan (A) and CT myelograms (B, C) showed abnormal translucent signals (white arrows) at the L3 spinal processes and laminae. Magnetic resonance images (D–F) showed an epidural mass lesion (white arrowheads) at the same level. This well-demarcated lesion seemed to be isolated from the intraosseous lesions (white arrows).

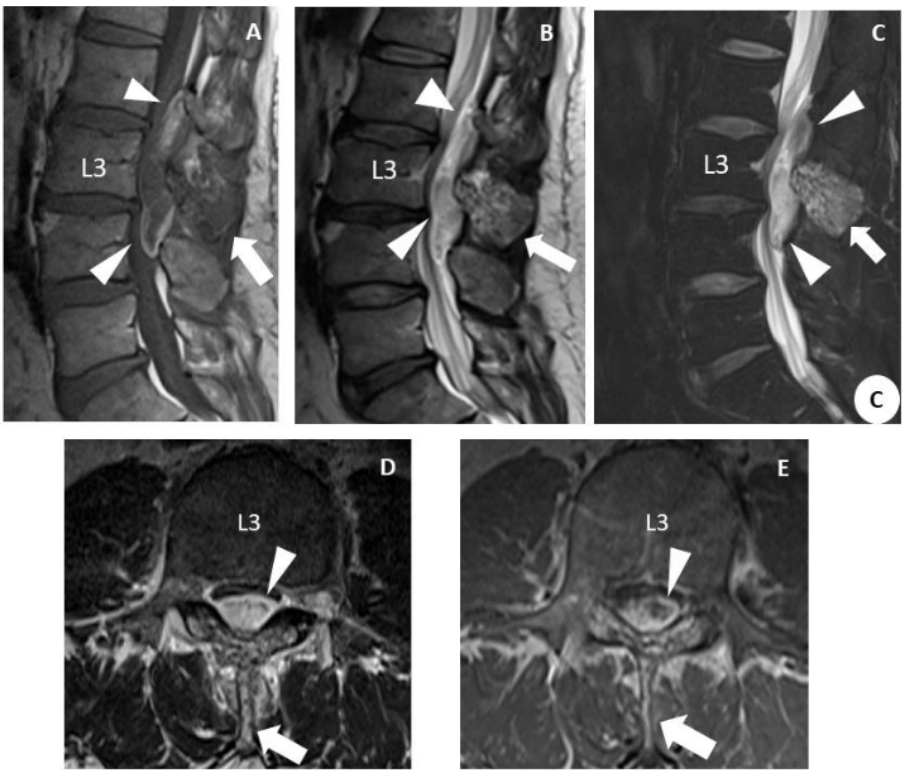

Figure 2 Preoperative sagittal (A-C) and axial (D, E) magnetic resonance images showed the rapidly enlarged epidural mass lesion (white arrowheads) that compressed the spinal cord anteriorly. The intraosseous lesions (white arrows) remained the same size.

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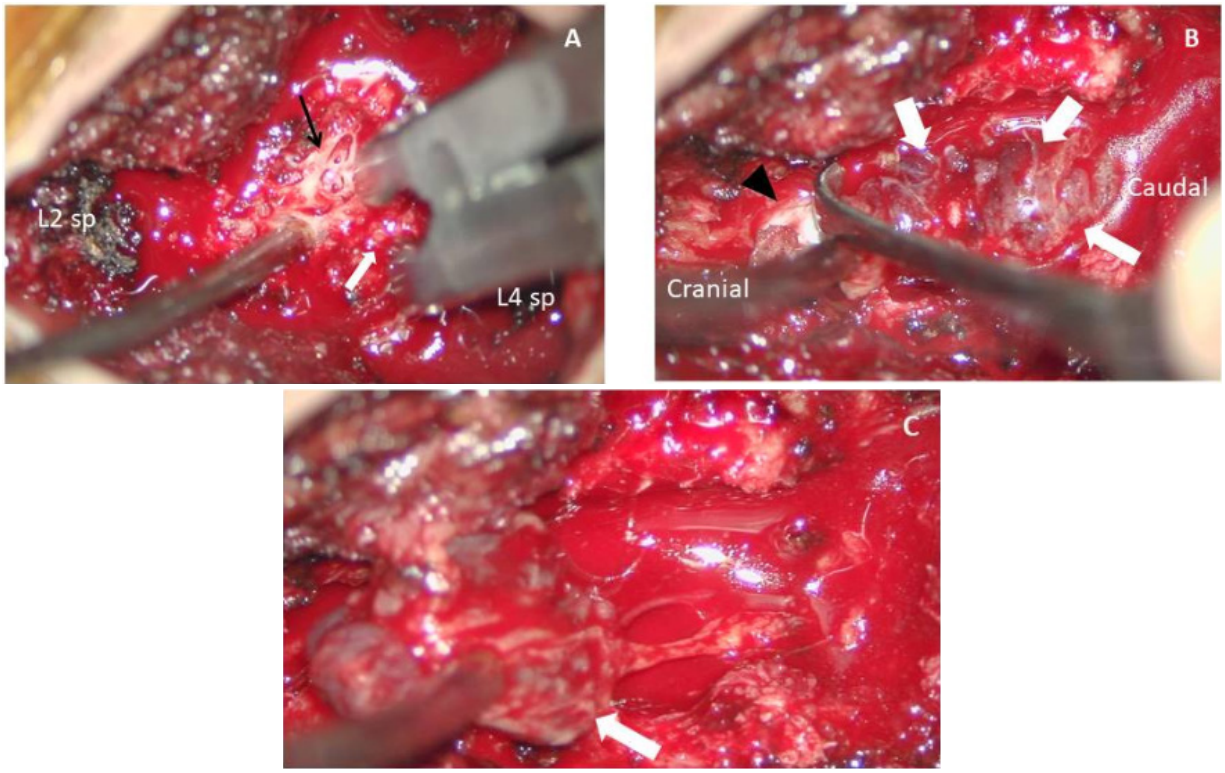

Figure 3 Intraoperative photographs. (A) Removal of the spinal processes and bilateral laminae at the L3 level exposed the soft and hemorrhagic tumor lesion (white arrows) in the apparently sparse trabecular bone (black arrow). (B, C) The reddish tumor lesion (white arrows) in the epidural space was macroscopically isolated from the bone lesions and covered with epidural fat tissue and a thin capsule. black arrowhead: dura mater.

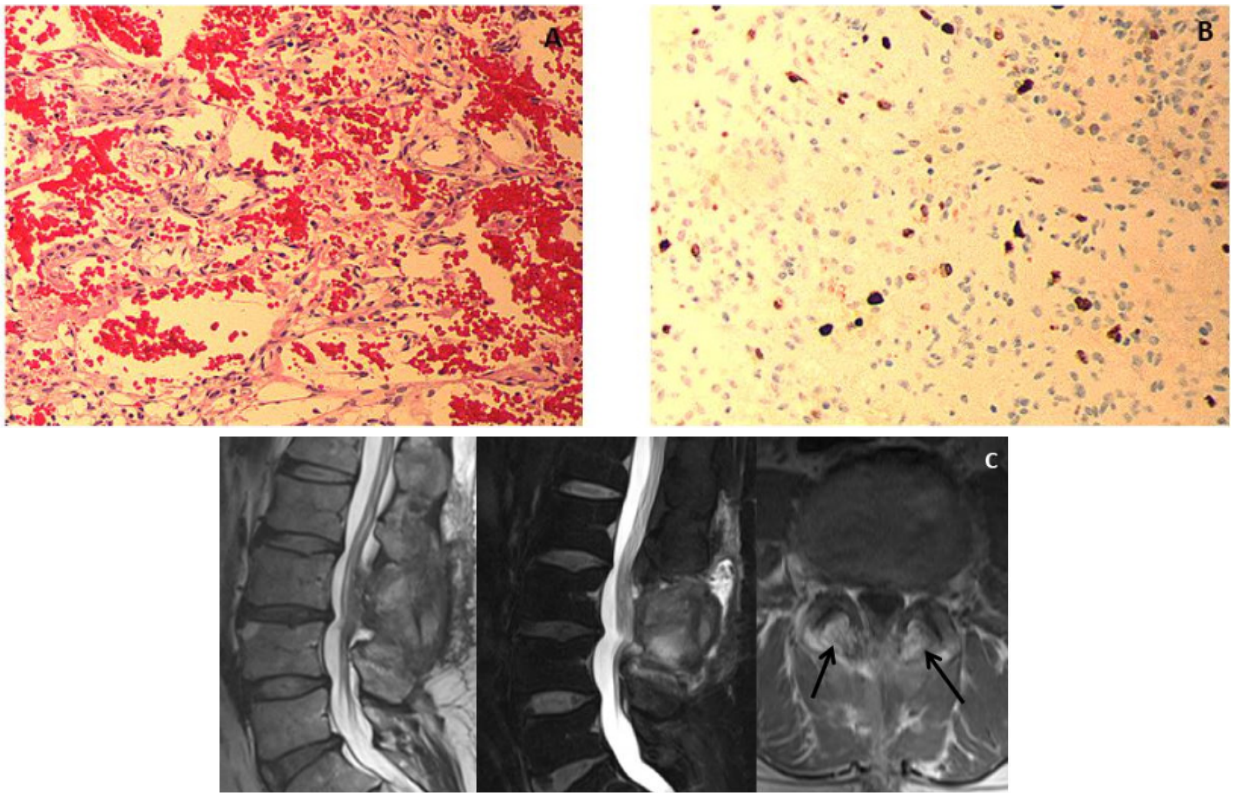

Figure 4 Histopathological findings. (A) Hematoxylin and eosin staining showed numerous capillary vessels of varying sizes intermingled with spindle-shaped lobular cells (×100). (B) MIB-1 index was not so high, approximately 2-3% (×100). These findings were compatible with capillary hemangioma. (C) Postoperative magnetic resonance images. The epidural mass lesion was completely resected, but abnormal intensity signals were apparent in the bilateral pedicles (black arrows), suggestive of remnant tumor.

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Discussion

The differential diagnosis of spinal epidural tumor lesions includes metastatic spinal tumor, meningioma, NST, lymphoma, lipoma, and hemangioma.[3,21] Most reported cases are metastatic spinal tumor and NST.[3,13] Hemangiomas are typically classified into cavernous, capillary, venous, and arteriovenous based on the histological findings.[13] Cavernous hemangioma is a congenital vascular malformation usually found in intracranial and subcutaneous tissues, and sometimes vertebral bodies,[3,15] whereas capillary hemangioma is often encountered in the skin, bone, and soft tissues in pediatric patients.[3,15] Histologically, blood vessels in cavernous hemangiomas are not separated by normal bone, but thin-walled capillary vessels in capillary hemangiomas were separated by normal bone.[22] Most spinal epidural hemangiomas are the cavernous type.[3] An online literature search of the PubMed database yielded 20 cases of SECHs (Table 1).[1–20] The 20 patients had a median age of 49.9±14.4 years and indicated a slight predilection for females with a male-to-female ratio of 8:12. The reported clinical manifestations were variable, and no case was detected incidentally. In particular, three cases occurred posttrauma. Most cases had foraminal extension, which allowed differentiation from cavernous hemangioma.[9] SECH tended to occur at the thoracic levels, and was rare in the lumbosacral regions as in our case. Complete resection was achieved without recurrence in most cases. No previous case of SECH was associated with isolated intraosseous lesion at the same level.

Table 1 Summary of previously reported cases of spinal epidural capillary hemangioma

Author and year	Age	Sex	Clinical symptoms	Spinal	Degree of tumor extension	Tumor resection	Recurrence
Akhaddar et al, 2010[1]	19	F	ND	T5-6	Foraminal extension	Complete	ND
Badinand et al, 2003[2]	40	F	GD, incontinence, and lower limbs pain	T2-4	Foraminal extension	Complete	None
Brasil et al, 2018[3]	69	F	Back pain	T9-10	None	Complete	None
Cofano et al, 2019[4]	52	F	ND	T6-9	Foraminal extension	Complete	ND
Egu et al, 2016[5]	60	F	S1 back pain, radicular pain	L5-S1	Foraminal extension	Complete	ND
García-Pallero et al, 2015	67	F	ND	T4-5	Foraminal and intrathoracic extension	Complete	ND
Gencpinar et al, 2014[7]	17	F	GD	T3-7	Foraminal extension	Complete	ND
Gupta et al, 1996[8]	50	M	ND	T8-10	Foraminal extension	Complete	None
Hasan et al, 2011[9]	57	M	Progressive myelopathy, low back pain	T10-12	Foraminal extension	Partial	None
Kang et al, 2006[10]	56	M	Chest wall pain	T2-4	Foraminal and intrathoracic extension	Partial	None
Kilic et al, 2017[11]	40	M	left leg pain, back pain (Posttraumatic)	L1	Vertebral body, pedicle, and transverse process	Complete	None
Niznick et al, 2020[12]	51	F	GD, sensory disturbance	T5-6	Foraminal extension	Complete	None
Rajeev et al, 2017[13]	50	M	Paraparesis, low back pain	T12-L2	Foraminal extension	Complete	None
Rajpal et al, 2020[14]	29	F	Flank pain	T7	Foraminal and intrathoracic extension	Complete	None
Seferi et al, 2014[15]	58	M	Paraparesis, sensory disturbance, and back pain	T2-4	Foraminal extension	Complete	None
Shilton et al, 2011[16]	47	F	Thoracic myelopathy, back pain (Posttraumatic)	T7-8	None	Complete	None
Sudhir et al, 2019[17]	63	M	Lower limbs weakness, back pain (Posttraumatic)	T6-8	Vertebral body, pedicle, and transverse process	Complete	None
Tekin et al, 2008[18]	56	F	L4 hypoesthesia, back pain	L3-4	None	Complete	None
Vassal et al, 2011[19]	59	F	ND	T5-7	Foraminal and intrathoracic extension	Complete	None
Xu et al, 2018[20]	57	M	ND	T2-3	Foraminal extension	Complete	ND
Current study	47	M	Sensory disturbance, incontinence, lower limbs pain, and back pain	L2-4	Spinal process and laminae	Partial	None

F, female; GD, gait disturbance; M, male; ND, not described

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Hereditary diseases such as *PTEN* hamartoma tumor syndrome or Klippel-Trenaunay-Weber syndrome sometimes cause multiple vertebral hemangiomas.[23,24] Vertebral cavernous hemangiomas extending outside the vertebral body are sometimes seen[24,25] but SECH occurred with intraosseous lesions in only two cases.[11,17] The epidural lesions in these cases were thought to result from adjacent bone destruction after spinal trauma. The pathogenesis of the tumor may involve release of local angiogenic factors or chronic repetitive irritation by microinstability of the posterior elements.[11,17]

Pathologically, capillary hemangioma is a benign vascular tumor lesion, but has high proliferative activity.[3] Rapid growth of capillary hemangioma in the skin or soft tissues is usually encountered, even within a few weeks or months.[21] On the other hand, rapid growth of capillary hemangioma in the central nervous system (CNS) is rare. Two causes have been proposed for rapid growth of capillary hemangioma. Firstly, underlying arteriovenous anastomosis with a small tumor remnant may be related to the recurrence of intradural capillary hemangioma after gross total resection.[21] A case of recurrent lobular capillary hemangioma on the skin was caused by angiogenic factors released from underlying arteriovenous anastomosis, which was confirmed by color Doppler study.[26] Anastomosis of SECH has not been proved, but the same mechanism may allow rapid growth. Secondly, increased secretion, intratumoral repeated hemorrhage, and inflammation may be related,[27] especially in the cases of ependymoma[28] and neuroenteric cyst.[27]

In our case, the SECH was not associated with inherited disorders, malignant tumors, or spinal trauma. Preoperative radiological examinations showed that the epidural lesion may have extended to form the intraosseous lesion without bony destruction. Both lesions were apparently isolated macroscopically, but were the same pathologically. No findings suggested inflammation or intratumoral hemorrhage. Therefore, we suggest that these tumors may have grown rapidly via underlying arteriovenous anastomoses. SECH seems to express the characteristics of vascular tumor more strongly than those of tumor with high proliferative activity.

SECH may be radiologically misdiagnosed as metastatic spinal tumor, meningioma, or NST. It must be aware that SECH sometimes grows rapidly in a short period, and that recurrence may occur even after total resection. Close clinical and radiological follow up is extremely important for SECH in spite of its benign character.

Conclusion

We report a rare case of SECH manifesting as epidural lesion associated with intraosseous lesion without continuous connection at the same level. This disease sometimes shows rapid growth or recurrence via underlying arteriovenous anastomosis. As in our case, progressive clinical symptoms indicate the possibility of tumor growth even if the lesion is well demarcated, which suggests of benign tumor. SECH in addition to metastatic spinal tumors should be considered in the differential diagnosis of spinal epidural tumors associated with intraosseous lesions.

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Conflicts of interest

None.

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