Epidural Hemangioma: A Clinical Series of Five Patients and Review of Literature for the Decade

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Abstract:

Background: Since 1929, only 171 epidural hemangioma cases have been reported. We report five epidural hemangiomas and review cases reported over the past decade in terms of radiological features and clinical findings.

Methods: Among patients operated on for spinal tumors at our hospital between 2009 and 2020, five had epidural hemangiomas. We retrospectively examined patient records and images and reviewed relevant English literature in PubMed from 2009 to 2019. Eighty-seven epidural hemangioma cases were reported in the last 10 years.

Results: Among 87 cases, the average age was 49.58 years; 43 and 44 cases were male and female, respectively. The most common lesion level was thoracic (59.8%), while common symptoms were back pain (42.5%); numbness, hypoesthesia, or anesthesia (37.9%); paraparesis (34.5%); and radicular pain (20.7%). On magnetic resonance imaging (MRI), 77.1% showed hypo-isointensity on T1-weighted image (WI), hyperintensity on T2WI, and homogenous enhanced patterns with contrast. Total resection was performed in most cases, with good clinical outcomes. Preoperative embolization was performed in four cases, with good surgical outcomes. The median follow-up duration of postoperative MRI was 16 months, and no case had recurrence.

Conclusions: Epidural hemangiomas are extremely rare and may be difficult to diagnose preoperatively. They should be considered if a dumbbell-shaped or epidural hypervascular lesion is noted. The treatment of choice for epidural hemangioma is total resection, with most cases having good clinical outcomes. Preoperative embolization may be useful for reducing bleeding. Although there were almost no recurrences after resection, careful follow-up for longer periods is required.

Keywords:

epidural tumor, hemangioma, magnetic resonance imaging

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Introduction

Hemangioma is the most frequent benign tumor involving the spinal column. According to autopsy findings, hemangiomas, originating from vertebral bodies, are seen in 11% of the general population[1]. In contrast, epidural hemangioma is extremely rare, accounting for only 4% of all extradural spinal tumors[2], which may be difficult to diagnose preoperatively. Only 171 epidural hemangiomas have been reported since 1929, of which 87 were noted within the last 10 years, including the 5 present cases. Long-term recurrence after surgery is especially rare, with only two cases, including one of our cases in the last 10 years. We report five epidural hemangiomas and analyze the cases reported over the past decade in terms of radiological features and clinical findings.

Materials and Methods

Among the patients operated on for spinal tumor at our hospital between January 2009 and February 2020, five cases were epidural hemangiomas. We examined the patient records and images retrospectively.

We examined available English literature on PubMed from January 2009 to December 2019. The following search terms were used in PubMed to obtain our studies: (“epidural” OR “extradural” OR “dumbbell”) AND (“hemangioma” OR “angioma”). A total of 87 epidural heman-

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Table 1. Characteristics and Symptoms of Our Five Patients.

Case	Age (years)	Sex	Clinical symptoms and signs	Duration of symptoms
1	70	Female	Dullness of both thighs; numbness on both soles	3 months (25 years after resection)
2	76	Female	Numbness on both soles; staggering when walking; lower extremity dullness; spasticity claudication	5 years, 3 months
3	75	Female	Left lower leg pain	2 years
4	66	Female	Numbness on the right lower leg; paraplegia	2 years, 6 months
5	49	Female	Lumbar discomfort; numbness and muscle weakness in the left lower leg	3 years, 2 months

Table 2. MRI Findings of Our Five Cases.

Case	Location level	T1WI	T2WI	Gadolinium-enhanced T1WI	Intervertebral foramen extension	Preoperative diagnosis
1	T7–T8	Hypo-isointense	Hyperintense	Homogeneous	Yes	Recurrence of hemangioma
2	T4–T5	Isointense	Hyperintense	Homogeneous	Yes	Meningioma, metastatic tumor
3	T12–L1	Isointense	Hyperintense	Homogeneous	Yes	Hemangiomas, meningiomas, schwannomas, neurofibromas, angiolipomas, lymphomas, metastases
4	T5	Isointense	Hyperintense	Homogeneous	Yes	Meningiomas, metastases, hemangiomas
5	L3	Isointense	Hyperintense	Homogeneous	Yes	Meningiomas, metastases, hemangiomas

Table 3. Surgical Results of Our Five Cases.

Case	Treatment	Preoperative embolization	Operation time (minutes)	Intraoperative bleeding (mL)	Histopathological diagnosis	Outcome	Postoperative MRI
1	Resection+radiation (30Gy/15Fr)	Yes	278	600	Mixed hemangioma (capillary, venous)	Completely improved	66 months-no recurrence
2	Total resection	Yes	238	170	Cavernous hemangioma	Completely improved	48 months-no recurrence
3	Total resection	No	170	300	Cavernous hemangioma	Good recovery	
4	Total resection	Yes	217	104	Cavernous hemangioma	Good recovery	90 months-no recurrence
5	Total resection	Yes	377	509	Mixed hemangioma (capillary, venous)	Good recovery	12 months-no recurrence

gioma cases have been reported in the last 10 years, including our 5 present cases.

Results

Table 1, 2, 3 list the patient backgrounds, symptoms, MRI findings, and surgical results of the five reported cases.

The average age was 67.2 years, and the median age was 70.0 years. All five patients were female. The most common symptom was numbness. One of the five cases had a recurrence 25 years after resection. The other four patients had symptoms for several years, of which three developed new symptoms in the previous few months (Table 1).

The levels of epidural hemangiomas were thoracic in three cases, thoracolumbar in one case, and lumbar in one case. MRI revealed isointensity on T1WIs and hyperintensity on T2WIs, with homogeneously hyperintense contrast enhancement in most cases. All cases had intervertebral foramen extension. The preoperative diagnosis included meningioma and metastasis (Table 2).

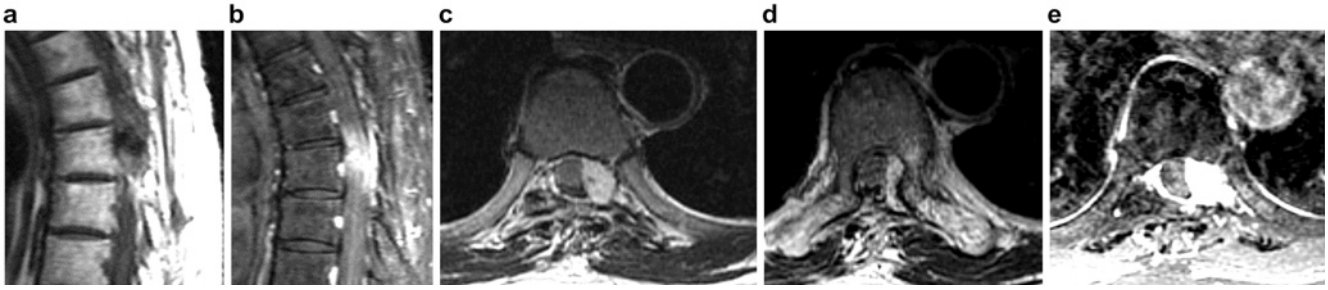

Figure 1. Preoperative magnetic resonance imaging (MRI) of case 1, 25 years ago.
(a) T1WI sagittal, (b) Gd-enhanced T1WI sagittal, (c, d) T2WI axial, (e) Gd-enhanced T1WI axial.

Resection and postoperative radiotherapy were performed in one patient, and total resection was performed in four patients. Preoperative embolization was performed in four of five patients. The average duration of operation was 256 (170-377) min, and the average intraoperative blood loss was 336.6 (104-600) mL. There were no postoperative complications, and the symptoms improved in all cases. In one case, recurrence was not evaluated because it occurred just after surgery. In the other cases, follow-up MRI was continued for 66 months; however, no recurrence was noted (Table 3).

Case presentations

Case 1

A 70-year-old woman presented with weakness in both thighs and numbness in both soles for 3 months. She was diagnosed with hemangioma and underwent resection at another hospital, 25 years prior to presentation. The symptoms improved after surgery. Similar symptoms gradually worsened starting 3 months prior to the present admission.

MRI 25 years ago revealed an extradural lesion extending from T7 to T8, which was hypo-isointense on T1WI and hyperintense on T2WI, with homogeneously hyperintense contrast enhancement. An axial image of the lesion documented its posterior/left lateral epidural location extending through the left T7-T8 neural foramen to the left rib process; the spinal cord was displaced to the right (Fig. 1). She was diagnosed with hemangioma and underwent resection. Subsequently, she had no symptom recurrence for 25 years, and follow-up MRI was not performed during that time. At presentation to our hospital, MRI showed the same findings; however, the lesions were slightly increased compared to those found 25 years ago (Fig. 2). The diagnosis was recurrence of epidural hemangioma.

Embolization was performed the day prior to surgery. The left eighth intercostal artery and the seventh and eighth right intercostal arteries were identified as the feeding arteries. Their main trunks were occluded. The day after the embolization, excision and T4-T11 posterior fusion were performed. The duration of operation was 278 min, and the intraoperative blood loss was 600 mL. The pathological diagnosis was mixed hemangioma. Complete excision was difficult because of scar tissue and adhesions with the dura and because incomplete excision was performed; thus, postoperative radiation therapy was performed at a total dose of 30 Gy/15 fractions.

After surgery and radiation treatment, the symptoms resolved completely, and there was no recurrence on MRI 66 months after surgery.

Case 2

A 76-year-old female suffered from numbness in both soles for 5 years, staggering when walking, lower extremity dullness, and spasticity claudication for 4 months. On physical examination, paresthesia was present bilaterally beyond the umbilicus.

MRI revealed an extradural lesion expanding from the T4 to T5 level, which was isointense on T1WI and hyperintense on T2WI, with homogeneously hyperintense contrast enhancement. An axial image revealed a left epidural dumbbell tumor extending through the left T4-T5 neural foramen to the left chest cavity (Fig. 3). The preoperative differential diagnosis was suspected meningioma or metastatic tumor.

Because it was suspected to be a hypervascular tumor, preoperative embolization was performed 2 days before the excision. The left fourth, fifth, and right fifth intercostal arteries, which were thought to be feeding arteries, were embolized (Fig. 4a, b). Two days after the embolization, excision and T3-T5 posterior fusion were performed. The duration of operation time 238 min, and the intraoperative blood loss was 170 mL. The pathological diagnosis was cavernous hemangioma (Fig. 4c). The numbness of the lower limbs gradually improved after the operation and had completely improved after 1 year. There was no recurrence on MRI 48 months after the surgery.

Case 3

A 75-year-old female suffered from left lower leg pain for 2 years. She limped and walked with a cane. There was pain from the left groin to the left thigh.

MRI revealed an extradural lesion expanding from the T 12 to L1 level, which was isointense on T1WI and hyperintense on T2WI, with homogeneously hyperintense contrast enhancement. An axial image revealed a right epidural dumbbell tumor extending from the spinal canal to the

Figure 2. Preoperative MRI of case1, present time.
(a) T1WI sagittal, (b) T2WI sagittal, (c) Gd-enhanced T1WI sagittal, (d) T2WI axial, (e) Gd-enhanced T1WI axial.

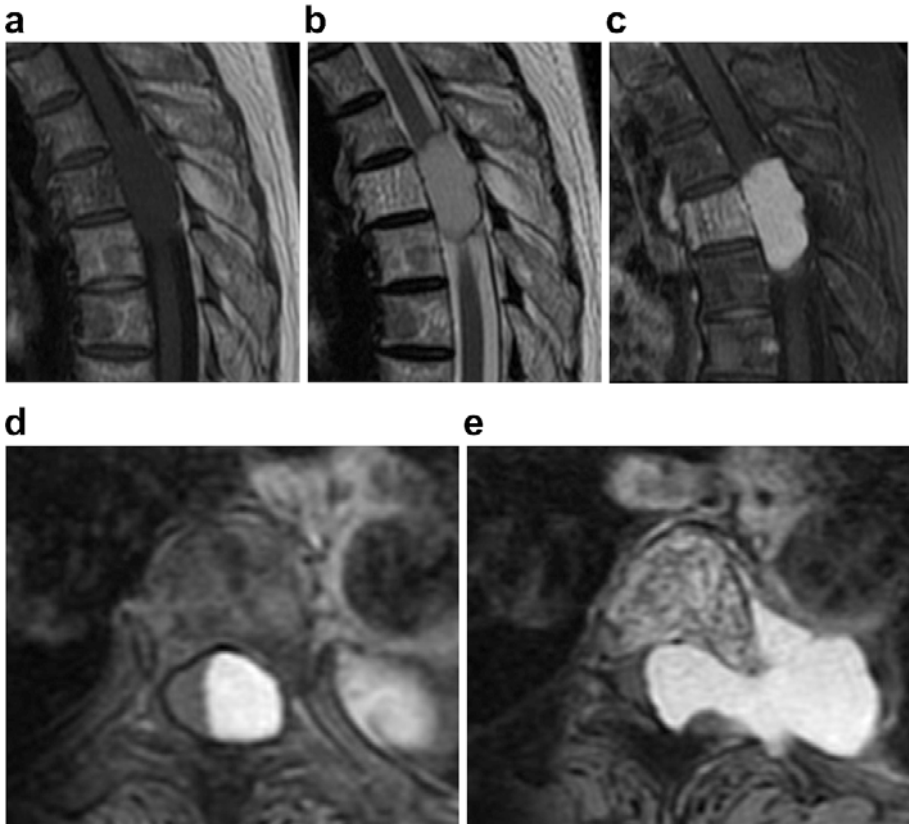

Figure 3. Preoperative MRI of case 2.
(a) T1WI sagittal, (b) T2WI sagittal, (c) Gd-enhanced T1WI sagittal, (d) T2WI axial, (e) Gd-enhanced T1WI axial.

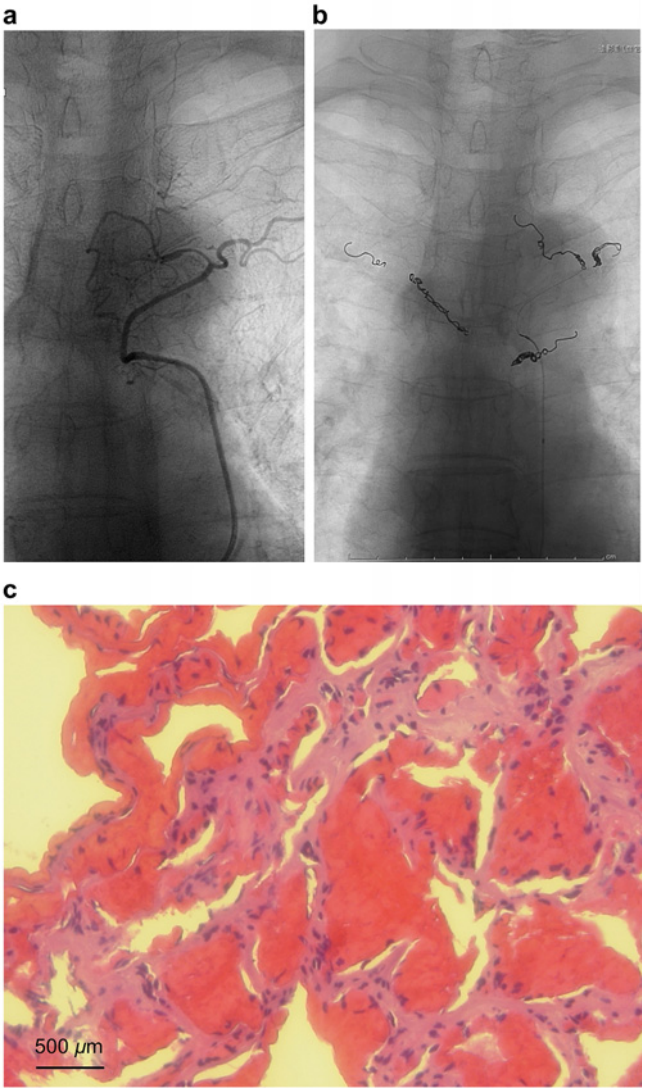

Figure 4. Angiographic image and pathological image of case 2.
(a) Angiographic image before preoperative embolization, (b) angiographic image after preoperative embolization, (c) hematoxylin and eosin staining.

retroperitoneum through the right T12-L1 neural foramen; the spinal cord was compressed to the right by the lesion (Fig. 5). Preoperative diagnosis included hemangiomas, meningiomas, schwannomas, neurofibromas, angiolipomas, lymphomas, and metastases.

Total resection and T12-L1 posterior fusion were performed. There was a red, easily hemorrhagic lesion on the right side behind the dural tube. The tissue was separated from the surrounding tissue and was excised en bloc. The duration of operation was 170 min, and the blood loss was 300 mL. The pathological diagnosis was cavernous hemangioma. The symptoms improved promptly after the operation.

Case 4

A 66-year-old female suffered from numbness on the right lower leg for 2 years and paraplegia for 6 months. Bilateral iliopsoas, quadriceps, tibialis anterior, and gastrocnemius muscles showed MMT4/4 level weakness. The patellar tendon and the Achilles tendon reflexes were enhanced bilaterally, and the Babinski sign was positive on both sides.

MRI revealed an extradural lesion at the T5 level, which was isointense on T1WI and hyperintense on T2WI, with homogeneously hyperintense contrast enhancement. An axial image revealed the lesion expanding from the T5 epidural region to the left foramen, and the spinal cord was displaced to the ventral side (Fig. 6). The differential diagnosis included meningioma, metastasis, and hemangioma; however, the preoperative diagnosis was difficult.

Total resection and T4-T7 posterior fusion were performed. The tumor was an epidural, dark red, vascular tumor extending from the ventral side of the spinal cord into the T5-T6 foramen (Fig. 7a). The tumor was completely removed. The duration of operation was 217 min, and the blood loss was 104 mL. The pathological diagnosis was cavernous hemangioma (Fig. 7b). The symptoms improved promptly after the operation. There was no recurrence on MRI 90 months after surgery.

Case 5

A 49-year-old female suffered from lumbar discomfort for 3 years and numbness and muscle weakness in the left lower leg for 2 months. Left iliopsoas, quadriceps, and tibialis anterior muscles showed MMT 3 level weakness. The left patellar tendon reflex and the both Achilles tendon reflexes were weakened.

MRI revealed an extradural dumbbell lesion at the L3 level, which was isointense on T1WI and hyperintense on T 2WI, with homogeneously hyperintense contrast enhancement. An axial image revealed an epidural lesion expanding to the left L2-L3 foramen, and the spinal cord was compressed to the right side. There was scalloping on the dorsal side of the L3 vertebra (Fig. 8). The differential diagnosis included meningioma, metastasis, and hemangioma. The preoperative diagnosis was hemangioma.

Embolization was performed the day before the extraction (Fig. 9). The feeding arteries were occluded from the left second lumbar trunk. Tumor staining was almost eliminated after the embolization.

The day after embolization, excision and L2-L4 posterior fusion were performed. The tumor was located in the epidural region on the dorsal side of the dural tube, involving the L2 and L3 nerve roots. The L2 nerve root was atrophied and adhered to the tissue around the tumor on the outer side. The tumor was completely removed. The duration of operation was 377 min, and the blood loss was 509 mL. The pathological diagnosis was mixed hemangioma.

The muscle strength improved several days after the operation, and numbness gradually improved. There was no recurrence on MRI 12 months after surgery.

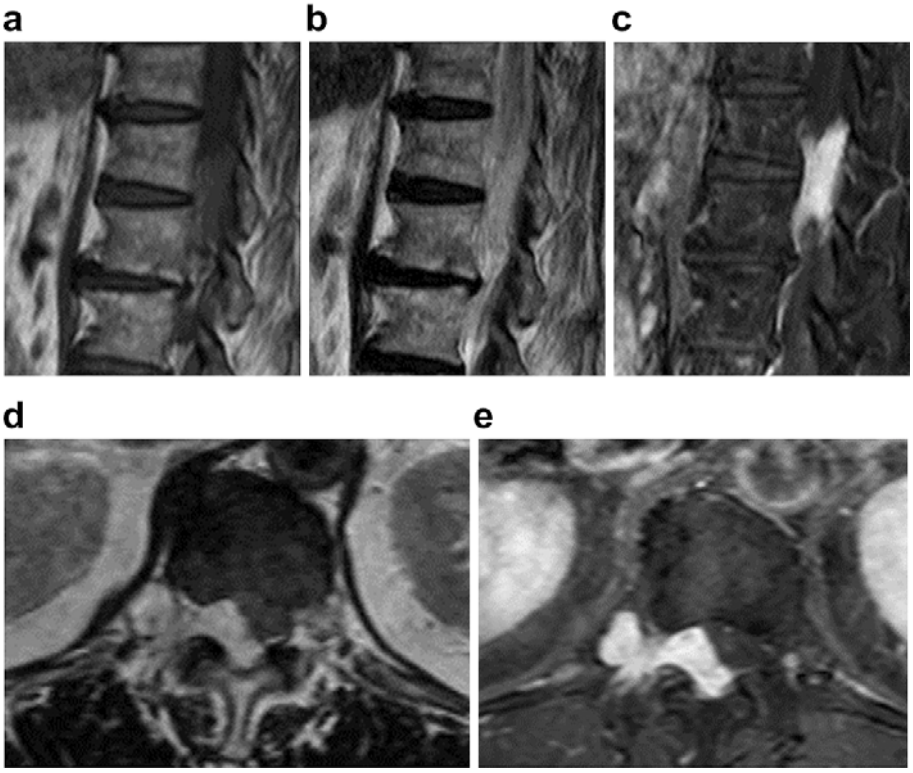

Figure 5. Preoperative MRI of case 3.
(a) T1WI sagittal, (b) T2WI sagittal, (c) Gd-enhanced T1WI sagittal, (d) T2WI axial, (e) Gd-enhanced T1WI axial.

Discussion

The majority of spinal hemangiomas originate from the vertebral bodies and may sometimes extend into the epidural space; they are usually asymptomatic lesions[2-4]. Epidural hemangiomas without vertebral body lesions are extremely rare and may be difficult to diagnose preoperatively. In a report of 678 primary spinal cord tumors in Japan, only 4.0% were hemangiomas, of which 11.1% were epidural[5]. Epidural hemangiomas have been reported in the English literature in only 171 cases dating back to 1929, with 87 cases reported in the last 10 years, including our 5 cases.

Patient background

Table 4 displays the summary of patient backgrounds, symptoms, lesion levels, and duration of symptoms of the 87 epidural hemangioma patients reported in the last 10 years. In our study, the average age was 49.58 years (9-78 years, median 51.0 years, standard deviation 16.0). There were 43 males and 44 females. Although it was previously reported that the number of cases was higher among male patients[6], no significant difference in the number of cases between male and female patients in the series was observed.

The most common lesion level was thoracic; there was 1 patient with a cervical lesion (1.1%), 6 with cervicothoracic lesions (6.9%), 52 with thoracic lesions (59.8%), 5 with thoracolumbar lesions (5.7%), 15 with lumbar lesions (17.2%), 3 with lumbar/sacral lesions (3.4%), and 5 with sacral lesions (5.7%). Consistent with previous reports[7], we found that epidural hemangiomas occur most often at the thoracic level, sometimes occur at the lumbar level, and rarely occur at the cervical level.

Symptoms

In our study of 87 cases, the most common symptom was back pain (42.5%). Numbness, hypoesthesia, anesthesia (37.9%), paraparesis (34.5%), and radicular pain (20.7%) were also common symptoms (Table 4). Hatiboglu et al. reviewed 80 cases of pure epidural cavernous hemangioma reported between 1929 and 2006. They found that 70% were female patients, and the peak age was around 40 years, with 19% hemangiomas extending into the neural foramen[7]. In their report, the main symptoms were local pain (4%), radiculopathy (10%), and paraparesis (86%)[7]. In our study and previous reports, the clinical manifestations of pure epidural hemangioma were back pain, paraplegia, and sensory deficits, mimicking disc herniation. In our study, at least 58.7% of cases on MRI showed progression to the neural foramen. The symptoms may be due to the progression to the neural foramen.

Of the 63 cases described in Table 4, there was variability in the duration of symptoms before surgery: 5 (7.9%) within 3 days, 7 (11.1%) from 3 days to 1 month, 26 (41.3%) from 1 to 6 months, 11 (17.5%) from 6 to 12 months, 11 (17.5%) for 1 year or more, and 3 (4.8%) recurrent cases. Of the 11

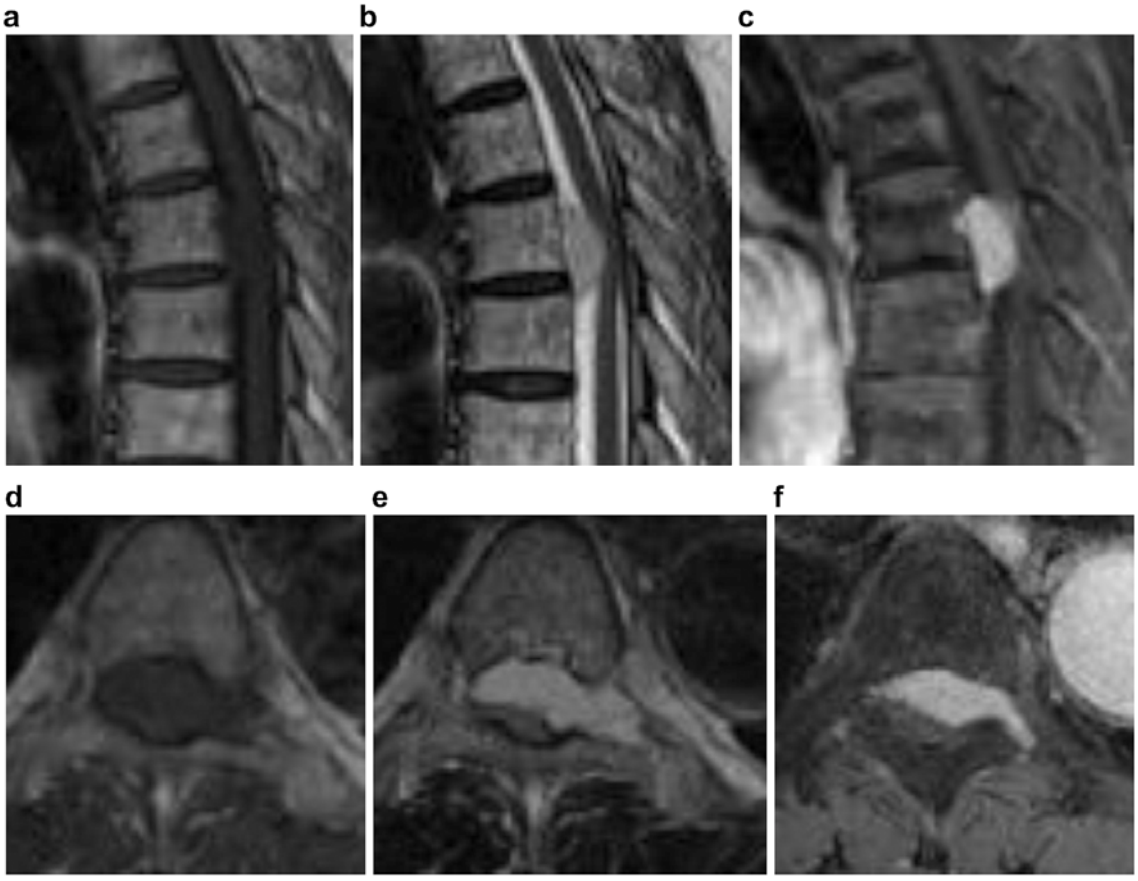

Figure 6. Preoperative MRI of case 4.
(a) T1WI sagittal, (b) T2WI sagittal, (c) Gd-enhanced T1WI sagittal, (d) T1WI axial, (e) T2WI axial, (f) Gd-enhanced T1WI axial.

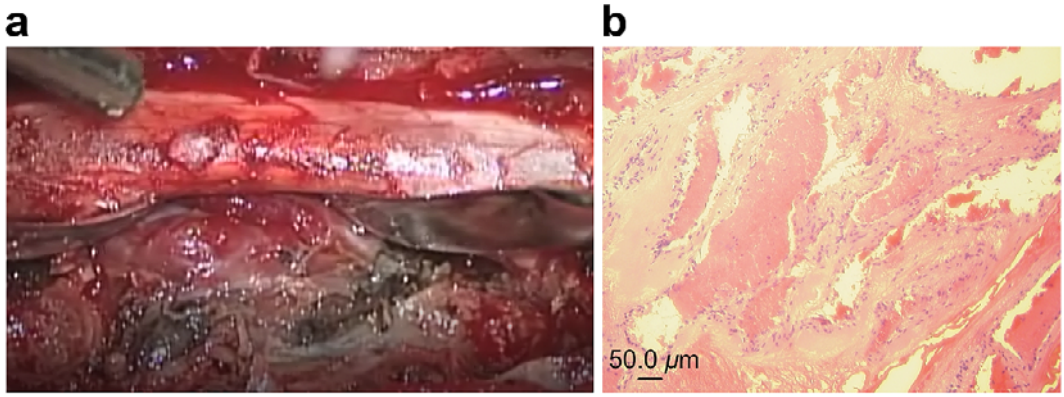

Figure 7. Intraoperative findings and pathological image of case 4.
(a) Intraoperative findings. There was a reddish vascular-rich lesion on the ventral side of the dura. (b) Hematoxylin and eosin staining.

patients with symptoms for more than a year, 5 had new symptoms within the final few months. In most cases, because epidural hemangiomas are benign lesions that grow slowly, neurological symptoms progress slowly over a month. However, because 7.9% of the cases had an acute onset within 3 days, it appears that neurological symptoms rarely develop acutely due to epidural hemorrhage and rapid increase of tumor resulting from microthrombus and bleeding within the tumor.

MRI findings

Table 5 summarizes 87 MRI findings in the last 10 years. On MRI, 77.1% (54/70) showed hypo-isointensity on TIWI, hyperintensity on T2WI, and homogenous enhance pattern on contrast. Contrast MRI revealed homogeneous enhancing patterns in 84.5% (60/71), heterogeneous enhancing patterns

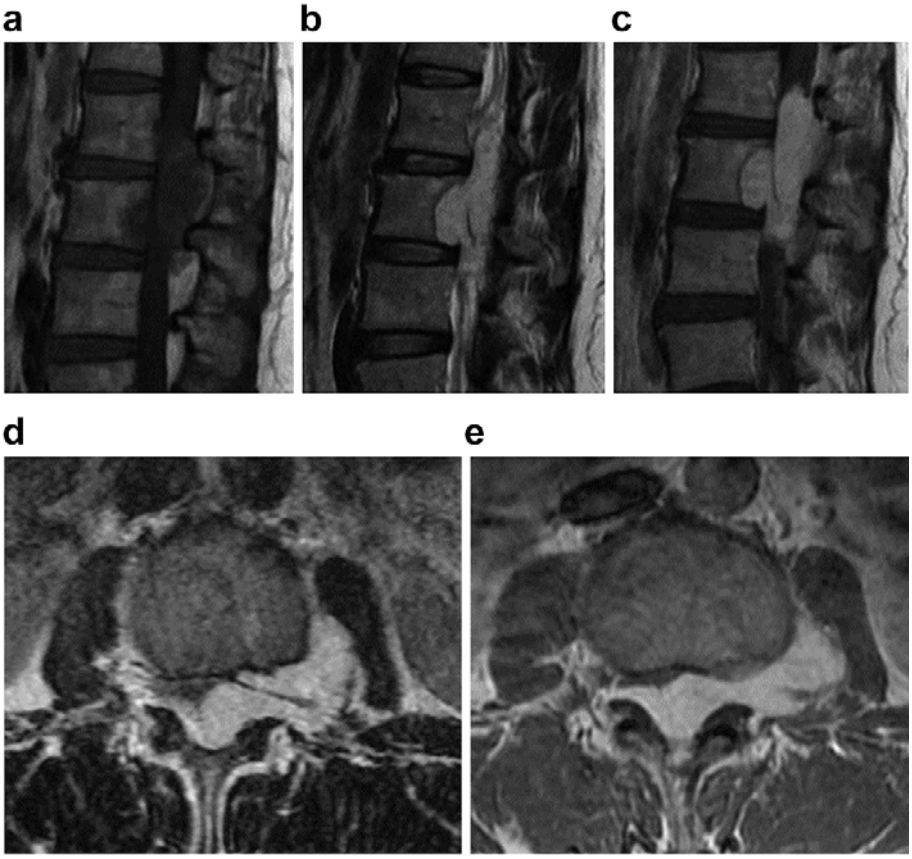

Figure 8. Preoperative MRI of case 5.
(a) T1WI sagittal, (b) T2WI sagittal, (c) Gd-enhanced T1WI sagittal, (d) T2WI axial, (e) Gd-enhanced T1WI axial.

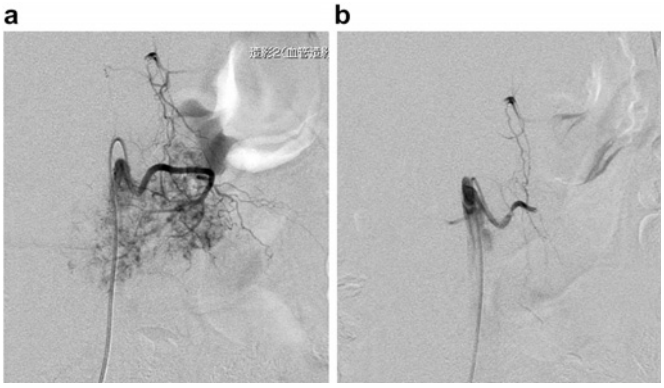

Figure 9. Angiographic image of case 5.
(a) Angiographic image before preoperative embolization. (b) Angiographic image after preoperative embolization.

in 11.3% (8/71), ring-enhancing patterns in 2.8% (2/71), and no enhancement in 1.4% (1/71).

As in previous reports[6], we found that in most epidural hemangiomas, MRI revealed hypo-isointensity on T1WI, hyperintensity on T2WI, and a homogeneous enhancement pattern with contrast. However, in our study, contrast MRI showed a heterogeneous enhancing pattern in eight cases, ring enhancement in two cases, and no enhancement in one case. Of the eight cases showing heterogeneous enhancing pattern, one case was an arteriovenous hemangioma[8], and one was a venous hemangioma[9]; the remaining six cases were all cavernous hemangioma. However, three patients have relatively acute onset of symptoms of 6 h[10], 3 days[11], and 8 days[10]. One of the two patients showing a ring-enhancing pattern had a cavernous hemangioma with acute onset within 48 h[11]. One case showing no enhancement was a cavernous hemangioma with acute onset within 48 h. Acute subdural hemorrhage was confirmed during surgery[12].

Our study suggests that contrast-enhanced MRI may show a heterogeneous enhancement pattern, no enhancement, or ring enhancement in cases with venous/arteriovenous hemangiomas, in the context of intralesional hemorrhage or acute onset. Nevertheless, because the numbers of cases of venous/arteriovenous hemangiomas and the cases of acute-onset epidural hemangiomas are very small, further study is needed.

In previous reports, it was reported that the number of cases that progressed to the neural foramen was relatively small. However, in our study, at least 57% (51/87) of cases showed progression to the neural foramen (Table 5).

Differential diagnosis

Differential diagnosis on MRI includes schwannoma, neurofibroma, meningioma, lymphoma, chordoma, metastasis, and disc fragments. Of the 87 cases examined, 46 had a preoperative diagnosis by MRI described in the paper. The

Table 4. Patient Summery of 87 Cases.

Sex		
Male	43 cases	49.4%
Female	44 cases	50.6%
Age (9–78 years old)		
Median age	51	years old
Average age	49.6	years old
Lesion level		
Cervical	1 case	1.1%
Cervical/thoracic	6 cases	6.9%
Thoracic	52 cases	59.8%
Thoracolumbar	5 cases	5.7%
Lumbar	15 cases	17.2%
Lumbar/sacral	3 cases	3.4%
Sacral	5 cases	5.7%
Clinical symptoms and sign		
Back pain	37 cases	42.5%
Hypoesthesia, anesthesia, numbness	33 cases	37.9%
Paraparesis	30 cases	34.5%
Radicular pain	18 cases	20.7%
Duration of symptoms		
~3 days	5 cases	7.9%
~1 month	7 cases	11.1%
~6 months	26 cases	41.3%
6~12 months	11 cases	17.5%
1 year~	11 cases	17.5%
Recurrence	3 cases	4.8%
NA	24 cases	-

Table 5. MRI Summary of 87 Cases.

T1-weighted image	
Hyperintense	3 cases
Isointense	56 cases
Hypo-isointense	6 cases
Hypointense	7 cases
Mixed signal	2 cases
NA	13 cases
T2-weighted image	
Hyperintense	66 cases
Hypointense	4 cases
Mixed signal	6 cases
NA	11 cases
Gadolinium-enhanced MRI	
Homogeneous enhancing pattern	60 cases
Heterogeneous enhancing pattern	8 cases
No enhancing pattern	1 case
Ring-enhancing pattern	2 cases
NA	16 cases
T1W1: hypo-iso, T2WI: hyper, Gd: homogeneous	
Yes	54 cases
No	16 cases
NA	17 cases
Intervertebral foramen extension	
Yes	51 cases
No	16 cases
NA	20 cases

preoperative differential diagnoses included 19 (41.3%) schwannomas, 15 (32.6%) meningiomas, and 9 (19.6%) neurofibromas. Only 13 cases (28.3%) listed hemangioma on the preoperative differential diagnosis, suggesting that purely epidural hemangioma may be difficult to diagnose in many cases. Epidural hemangiomas should also be suspected if there is a dumbbell-shaped or epidural hypervascular lesion.

Pathology

Hemangiomas are congenital vascular malformations. Hemangiomas that occur in the spinal cord are classified by the predominant type of vascular channel (capillary, cavernous, arteriovenous, or venous) observed on histological examination.

In our study of 87 cases, cavernous hemangioma was most common, which occurred in 68 patients (81.0%), while capillary hemangioma occurred in 8 patients (9.5%), venous hemangioma occurred in 1 patient (1.2%), arteriovenous hemangioma occurred in 1 patient (1.2%), and mixed hemangioma occurred in 6 patients (7.1%) (Table 6). Consistent with other reports, we found that cavernous hemangioma was most common. Of the 87 cases, regardless of the type of hemangioma, most were successfully recovered by excision, and there is likely no difference in surgical outcome based on histology. However, there are few reports of hemangiomas other than the cavernous type; therefore, further study is necessary.

Treatment

The treatment of choice for epidural hemangioma is total removal of the lesion. In some reports, intraoperative findings reported almost no adhesions with the dura or surrounding tissues[13-15]; total resection was performed in the majority of the 87 cases over the past 10 years (Table 6). Some cases reported strong adhesion with the nerve roots and surrounding tissues[16,17], and subtotal resection was performed in some cases. Although subtotal resection was performed, one case was reported with symptoms completely resolving after surgery, and no exacerbations or recurrences were observed on MRI 16 months after surgery[10].

In the study of 87 cases over the last decade, radiation therapy was performed in 3 cases (Table 6). One patient underwent radiosurgery for the recurrent hemangioma several months after the subtotal resection, with symptoms completely resolving after radiosurgery. MRI 36 months after the surgery documented the regression and effective control[18]. Subtotal resection with postoperative radiation therapy was performed for two patients, including our "case 1"[19]. Our "case 1" is a case of recurrence 25 years after excision; the symptoms resolved completely after surgery and radiotherapy, and no recurrence was observed on MRI 66 months after surgery. Postoperative radiation therapy remains controversial[7]. It was performed for patients undergoing incomplete resection, and their reported results were good, sug-

Table 6. Summary of Treatment Results for 87 Cases.

Treatment	
Total resection	49 cases
Subtotal resection	2 cases
Resection (unknown)	31 cases
Subtotal resection+radiotherapy	2 cases
Radiosurgery	1 case
Biopsy	1 case
NA	1 case
Preoperative embolization	
Yes	5 cases
No	1 case
NA	81 cases
Histopathological diagnosis	
Cavernous hemangioma	68 cases
Capillary hemangioma	8 cases
Venous hemangioma	1 case
Arteriovenous hemangioma	1 case
Mixed hemangioma	6 cases
NA	3 cases
Postoperative course	
Completely improved	26 cases
Partially improved	1 case
Good recovery	34 cases
No change	1 case
Originally no symptom	2 cases
NA	17 cases

gesting that radiation therapy is recommended after surgery[20]. In contrast, there are reports that postoperative radiation therapy was not appropriate, even in a case of subtotal resection[21].

Of the 87 cases examined, only 5[22], including 4 of the 5 presented here, underwent preoperative embolization (Table 6). In our series, all of the four cases showed strong enhancement even in the arterial phase of the contrast image and were considered to be pleomorphic tumors. It was expected that the amount of bleeding during the operation would be large. All cases that involved preoperative embolization showed good surgical outcomes. Preoperative embolization can be expected to reduce the tumor volume and suppress bleeding. However, in the past 10 years, only five cases of preoperative embolization have been reported; therefore, it is necessary to collect more data in the future.

In our "case 1" and "case 2," hemangiomas were also found in the vertebral body. Vertebral hemangiomas occur in approximately 11% of the general population according to autopsy findings[1]; however, only 0.9-1.2% of all vertebral hemangiomas are symptomatic, and aggressive vertebral hemangiomas causing spinal cord compression are rare[23]. Total resection is effective for the treatment of aggressive vertebral hemangioma[23]. Radiation therapy has been used most often to treat local pain[24]. Some recommend additional radiation therapy after total excision or decompressive surgery[25]. In these two cases ("case 1" and "case 2"), the neurological symptoms improved, and no pain was reported after the extra-vertebral lesion was removed; therefore, radiation therapy for vertebral body was not performed, and there were no recurrences thereafter.

Postoperative recurrence

Regardless of which surgical method or radiotherapy for epidural hemangioma was used, good results were obtained postoperatively in almost all cases (Table 6). During the follow-up period of MRI with a median of 16 months (1-144 months, mean 31.2 months, standard deviation 31.7), no postoperative recurrences were observed, including recurrent surgery cases. However, of the 87 cases reported, 3 were postoperative recurrences. In these three cases, the symptoms recurred several months after the partial excision[18], 12 years after the resection[26], and 25 years after the resection (our present "case 1"). Therefore, although it was reported that there are almost no recurrences after resection of epidural hemangiomas, the present study suggests that careful follow-up for longer periods is required because the lesion may recur 10 years or more after surgery.

Conclusions

Epidural hemangiomas are extremely rare and may be difficult to diagnose preoperatively. The clinical manifestations are back pain, paraplegia, and sensory deficits. In most cases, neurological symptoms progress slowly over a month, but they rarely developed acutely due to epidural hemorrhage or rapid increase in tumor size. In most epidural hemangioma cases, MRI showed hypo-isointensity on T1WI, hyperintensity on T2WI, and a homogeneous enhancement pattern. Epidural hemangiomas should be considered if there is a dumbbell-shaped or epidural hypervascular lesion, although this presentation is easily confused with those of meningioma, schwannoma, neurofibroma, and disc herniation. The first choice of treatment for epidural hemangioma is total resection, with most patients showing good recovery after surgery. Preoperative embolization may be useful for reducing the amount of bleeding. Although it has been reported that there are almost no recurrences after resection, careful follow-up for longer periods is required.

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Ethical Approval: None.

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