Clinical Study

Clinical and imaging findings in patients with aggressive spinal hemangioma requiring surgical treatment

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Abstract

Vertebral hemangiomas (VHs) are frequently asymptomatic lesions found incidentally during investigations for other spinal problems. Symptomatic VHs are less common, and there are few reports of compressive VHs in the literature. VHs with aggressive behavior present with low signal intensity on T1-weighted and high signal intensity on T2-weighted MRI. We present a case series of four patients with compressive VH, all of whom were neurologically compromised. Each of the four patients underwent preoperative arterial embolization followed by surgical treatment of their VHs. All patients recovered normal motor function after surgery. At follow-up (average 53 months), one patient had a recurrent tumor requiring reoperation and radiotherapy. Although it is rare, aggressive VH can be a devastating condition.

Total surgical resection or subtotal resection with radiotherapy may be warranted.

Keywords:
Cord compression
Vertebral hemangioma
Spinal tumor

1. Introduction

Vertebral hemangiomas (VHs) are dysplastic tissues or vascular tumors that do not usually display malignant behavior.\textsuperscript{1} Autopsy and imaging studies have shown a VH incidence of 10–12%.\textsuperscript{2–4} The thoracic spine is the most common segment affected by VHs, but up to 30% of lesions involve multiple levels.\textsuperscript{3} Most VHs are asymptomatic and are diagnosed incidentally during investigations for other spinal problems,\textsuperscript{5} but infrequently patients with VH can present with pain or symptoms of neurological compression.\textsuperscript{2,5–12} VHs can be classified as asymptomatic, symptomatic or compressive based on their behavior and their radiologic appearance on plain radiography, CT and MRI.\textsuperscript{4,13}

The imaging characteristics of VHs are reliable indicators of the aggressiveness of the lesion. VHs that compress neural elements characteristically present as soft tissue on CT scans and have low signal intensity on T1-weighted and high signal intensity on T2-weighted MRI.\textsuperscript{4,13}

There is no consensus on the best therapeutic option for compressive VHs. Previous publications have reported only a limited number of cases and have mixed patients with symptomatic VHs producing back pain alone with cases of compressive hemangioma.\textsuperscript{3,5,8} Several treatment alternatives for compressive VH have been used, including arterial embolization,\textsuperscript{14} intrallesional ethanol injection,\textsuperscript{15,16} laminectomy with or without fusion, corpectomy and anterior column reconstruction, vertebroplasty\textsuperscript{17} and radiotherapy (either alone\textsuperscript{8,19} or as an adjuvant therapy).

Here we report a series of four patients with radiologically and biologically confirmed compressive VH who were treated with embolization and surgery. The patients had a median follow-up of 53 months.

2. Patients and methods

In a retrospective review of patient records from between 1998 and 2008, we identified four patients (three males, one female) with a median age of 50.5 years (range 14–72 years) who had undergone surgery for compressive VH. All patients presented with progressive neurological deficits but no other symptoms. The institutional review board approved the protocol used in this study.

Inclusion criteria were: (1) the presence of a VH with dural sac compression; and (2) a low-intensity signal on T1-weighted MRI but a high-intensity signal on T2-weighted MRI (Fig. 1). Preoperative investigations included a complete neurological assessment, plain radiography and MRI. In every case, the diagnosis was made based on the MRI characteristics combined with the observation of thecal sac compression. The level of the bone lesion, the site of dural compression and the results of the preoperative neurological assessment were recorded.

All patients underwent preoperative arterial embolization of their VHs. The surgical procedure was then performed, and any perioperative complications were recorded. Patients were first seen in the clinic 1 week after discharge from the hospital, then at 1, 3, 6 and 12 months after surgery, and then yearly thereafter. At each visit, a complete neurological examination was performed, and plain radiographs were obtained. A CT scan was obtained for every patient at 1 year after surgery. Any instances of recurring
neurological compression by the VH, reoperations and indications for radiotherapy were also recorded.

3. Results

For all patients, only one vertebra was involved in each case. Three VHS were located in the thoracic spine, and one was in the lumbar spine. All patients presented with paraparesis associated with dural sac compression. Two patients experienced anterior compression, with the tumor originating in the vertebral body and extending to the anterior aspect of the spinal canal, whereas the other two patients had circumferential involvement, with the VH again originating in the vertebral body and involving the entire canal (Table 1). Three patients presented with neurological involvement of less than 48 hours’ duration, while the fourth patient presented with a 2-week history of progressive paraparesis (Table 1).

3.1. Surgical procedure

All patients underwent arterial embolization, which was performed by an interventional radiologist before surgery (Fig. 2); no complications were observed as a result of this procedure. Interestingly, a patient with T5 involvement and anterior compression experienced complete neurological recovery after embolization.

Embolization was followed by surgery in all patients. Two patients with circumferential compromise of the thoracic spine underwent a posterior approach including laminectomy, tumor removal from the canal and instrumented fusion. One of the two, who had T6 involvement, also underwent open vertebroplasty because we judged that tumor removal would compromise the stability of the vertebral body. The only patient with lumbar (L5) VH that caused anterior compression was also treated with laminectomy, tumor removal from the canal and posterior instrumented fusion. One patient with only thoracic anterior compression underwent transthoracic corpectomy of T5 with a anterior instrumented fusion with a strut allograft (Fig. 3). This patient was the only one in whom complete macroscopic removal of the VH was achieved through surgery.

For all four patients, no perioperative complications were seen, and transfusions were not required.

3.2. Follow-up

Neurological assessments revealed complete motor recovery during the first week after surgery for all patients with thoracic involvement (patients 1, 2 and 4). The patient with a lumbar VH had regained normal motor function by 1 month after surgery.

Pathological examinations revealed cavernous hemangioma in two patients, and the other two patients were diagnosed with hemangioma (Table 1).

Although radiotherapy was indicated after surgery in all three patients with incomplete macroscopic removal of the lesion, two of these patients refused radiotherapy.

Follow-up ranged from 40 to 72 months (mean 53 months). Neither the recurrence of neurological compromise nor regrowth of the hemangioma was observed in three patients, namely the patient who underwent radiotherapy, the patient with complete macroscopic tumor removal, and the patient with lumbar spine involvement, who did not receive radiation or undergo complete removal of the VH. This third patient, patient 3, was followed postoperatively for 72 months until she died of causes unrelated to the VH, and did not experience any clinical or radiological recurrence during that time. One patient with a VH in the thoracic spine, who refused radiation, experienced recurrence of neurological

![Fig. 1. Sagittal MRI of the thoracic spine showing (A) compression of the spinal cord by a T12 hemangioma with a low-intensity signal on T1-weighted MRI and (B) a high-intensity signal on T2-weighted MRI.](image-url)

### Table 1

<table>
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<th>Patient</th>
<th>Age (years)</th>
<th>Sex</th>
<th>Vertebral level</th>
<th>Dural compression</th>
<th>Surgery</th>
<th>Radiotherapy</th>
<th>Follow-up (months)</th>
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<td>47</td>
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</table>
symptoms as well as regrowth of the VH into the spinal canal, with anterior compression of the cord, at 15 months after surgery (Table 1). At that time, the patient again underwent embolization in addition to anterior corpectomy and anterior fusion with an expandable cage (Fig. 4). The patient recovered normal motor function the day after the second procedure. He again refused radiotherapy, but no recurrence was observed in the 25 months of follow-up after his second operation.

4. Discussion

Symptomatic hemangiomas have been frequently reported in the literature; however, compressive hemangiomas are less common. Here, we present a series of four patients with compressive VHs. Each patient had a soft mass in the spinal canal and MRI findings of aggressive behavior. Patients had a mean follow-up of 51 months.

All cases of symptomatic VH had low-intensity signals on T1-weighted and high-intensity signals on T2-weighted MRI; these
characteristics differ from those of the usual asymptomatic VHs, which present with high-intensity signals on both T1- and T2-weighted MRI. Low signal intensity on T1-weighted MRI has been associated with a more active vascular lesion.

Neurological symptoms can result from tumor growth creating an extradural mass. This growth, in turn, produces compression, acute hemorrhage into the epidural space or spinal cord ischemia because of hemodynamic effects such as arterial “steal” and venous hypertension. Limited information is available in the literature on this hemodynamic effect; however, we noted that one of our patients experienced neurological recovery immediately after embolization, which is consistent with the findings of an earlier study, in which embolization alone was found to be successful at reducing neurological symptoms. Preoperative embolization has also been reported to reduce intraoperative bleeding and improve surgical field visualization. Some authors have shown that performing surgery in VH patients without first performing embolization can result in very significant blood loss. In our patients, preoperative embolization allowed adequate visualization of the surgical field, and none of our patients required blood transfusion during or after surgery; however, we did not have a control group that was operated on without preoperative arterial embolization, against which to compare blood loss. Preoperative arterial embolization is also recommended before surgery because the angiogram can also detect major spinal cord vessels in the area to be operated on, thus reducing the risk of spinal cord ischemia related to surgery.

Intralesional injections of ethanol have been used to relieve cord compression. We did not perform this procedure because devastating neurological complications and subsequent pathological fractures have been reported. Surgical treatment via an anterior or posterior exposure is currently advocated to treat progressive neurological deficit. Consistent with findings from other reports, the results of our study show that surgery can be performed safely in patients with compressive VH. Posterior decompression with or without instrumented fusion is recommended in patients with total vertebral involvement and circumferential cord compression. Decompression is also recommended in patients with rapid and progressive neurological compromise, such as our patients with circumferential compression and acute neurological compromise. An anterior corpectomy and reconstruction is preferred for patients with vertebral body involvement alone and for patients with more extensive vertebral involvement but anterior compression alone. One of our patients was operated on using an anterior approach, which allowed complete VH removal without subsequent recurrence and successful anterior column reconstruction (Fig. 3).

Radiation therapy has been used either as the sole treatment for VH or as an adjuvant therapy, primarily after subtotal VH removal. Most reports recommend radiation as an adjuvant therapy, particularly if only subtotal removal of the lesion has been achieved; conversely, radiotherapy does not seem to be necessary if the lesion is completely removed.

In patients with compressive VH, surgical treatment should be performed and should be followed by radiotherapy in the case of subtotal resection.

References