Management of aggressive vertebral hemangioma with cord compression

Ehsanali Alibai MD, Mosa Taghipour MD, Golnaz Yadollahi MD

Shiraz Neurosciences Research Center, Neurosurgery Department, Shiraz University of Medical Sciences, Shiraz, Iran

Address correspondence to: Dr. Ehsanali Alibai, Shiraz Neurosciences Research Center, Chamran Hospital, Chamran Boulevard, Shiraz, Iran. PO Box: 7194815644, Tel: +98711-6234508, Fax: +98711-6234508, E-mail: alibaiehsanali@yahoo.com, neuroscien@sums.ac.ir

Abstract

Objective: Aggressive vertebral hemangiomas (VHs) causing spinal compression are rare and there is controversy regarding treatment. This study aims to evaluate clinical results of patients with aggressive VHs after laminectomy, radiotherapy and vertebroplasty with spinal fixation and to discuss treatment options of tumors. Methods: We performed a retrospective study in 8 patients with aggressive VHs treated with laminectomy, radiotherapy and vertebroplasty with spinal fixation. In all the patients, tumor was either in thoracic or lumbar spine resulting in myelopathy with extraosseous extension. Tumors were assessed using magnetic resonance imaging (MRI) and the clinical results were evaluated. Results: All of the tumors showed low-intensity or low to isointensity signal on T1-weighted MRI. Laminectomy with or without irradiation was performed in 5 patients. Two patients underwent vertebroplasty with spinal fixation and conventional radiotherapy was performed in 1 patient. There was no preoperative complication. The myelopathy and patients' symptoms improved after the surgery. None of the patients had a recurrence at a mean follow-up period of 48 months. Conclusions: A combination of laminectomy, radiotherapy and vertebroplasty with spinal fixation for aggressive VHs with extraosseous extension caused spinal cord compression in all the patients. The clinical results proved satisfactorily in the long-term follow-up.

INTRODUCTION

Vertebral hemangiomas (VHs) are the most commonly encountered tumor of vertebral column which are mostly asymptomatic and very slow-growing.1 They may be detected as incidental roentgenographic findings or when they produce local pain. Only 0.9% to 1.2% of the VHs become symptomatic in some part of life.2 VHs which 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.3 The incidence of VHs as the most common benign spinal neoplasms has been differently reported from 10 to 27% based on autopsy series, plain X-rays and MRI reviews.4 The age distribution peaks between the third and fifth decades and there is a slight predominance in females, with a female-to-male ratio of about 2:1. Predominant anatomical sites are thoracic and upper lumbar spine.5 Three different histological types are capillary, cavernous, and mixed VH.6 The incidence of VH on spinal MRI in Northern Iran was 26.9%. It was more common in females (30%) than males (23%) in older age group and in lumbar spine.4 We performed surgery and radiotherapy on 8
patients to remove aggressive VHs with extraosseous extension causing spinal cord compression and neurological symptoms.

METHODS

We defined aggressive VHs as those with extraosseous extension that are causing spinal cord compression and neurological symptoms. We performed a retrospective study in which 8 patients, aged 22 to 48 years, with VH in different degrees of spinal cord or nerve root compression and without any prior therapeutic management were eligible to participate. The patients were enrolled from 2004 to 2010 to undergo surgery in Chamran Hospital, affiliated to Shiraz University of Medical Sciences. Written informed consent was obtained from all patients. The surgeries were carried out in 7 patients and 1 patient was treated with a fixed dose of external beam radiotherapy. In 6 patients, the vertebral hemangioma was in the thoracic spine and in the remaining 2 patients, it was in the lumbar spine. The tumor had an extraosseous extension that was causing myelopathy. The mean follow-up period was 48 months.

RESULTS

Patient characteristics are shown in Table 1. Three patients out of 8 were diagnosed by MRI. In 2 patients, diagnosis was confirmed by CT scan showing typical 'polka dot' appearance. In all the patients, spinal cord compression occurred due to extension of tumor into the extra dural space. In one patient, there was also expansion of the involved vertebra. After diagnosis was reached, the surgical procedure was performed; the posterior laminectomy with adjuvant radiotherapy was then performed in 5 patients. One patient was only treated by conventional radiotherapy. Vertebroplasty with spinal fixation was performed in the remaining 2 patients. Patients were followed at one week after they were discharged from hospital, then at 1, 3, 6 and 12 months after surgery, and yearly thereafter. At each visit, a complete neurological examination was performed and plain radiographs were obtained. CT scan was obtained from each patient at one year after the surgery. Any instances of recurring neurological compression by the VH, reoperations and indications for radiotherapy were recorded. The 6-month follow-up period showed good recovery of symptoms in almost all of the patients.

DISCUSSION

Hemangiomas are benign slow growing vascular tumors composed of newly formed capillary, cavernous or venous blood vessels. Among skeletal locations, vertebrae are second most common site. Incidentally, most VHs, solitary and asymptomatic, are often discovered radiographically. However, local pain, radiological aggressiveness and neurologic deficits may be present. In this study, we presented a series of 8 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 6 months. Management of symptomatic VHs that was reported in other studies was also mentioned (Table 2). The initial complaints of the patients with symptomatic VHs were localized pain and gradual weakness of lower extremities. Neurologic complications may dominate the clinical picture with compression of the nerve root, spinal cord or cauda equina. Symptomatic hemangiomas of the vertebral bodies associated with neurologic manifestations are usually located in the mid thoracic region, where diameter of the spinal canal is also small. Cord compression is more likely to occur with lesions that extend into the pedicles and laminae of thoracic vertebra where cord
occupies most volume of spinal canal.²²,²³ Sometimes, cord compression may occur due to 1) expansion of the involved and therefore enlarged, deformed vertebra encroaching upon the spinal cord, 2) extension of the tumor into the extra dural space, 3) extra dural hematoma, and 4) rarely because of compression fracture of the involved vertebra.²⁴ In all of our patients, spinal cord compression occurred due to extension of tumor into the extra dural space. In one patient, it was also due to expansion of the involved vertebra.

The imaging characteristics of VHs are reliable indicators of 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. All of our 8 VHs with extraosseous extension causing spinal cord compression showed low intensity or low-isointensity signals on T1-weighted MRI.¹⁴

There are several options for treating the aggressive VHs with extraosseous extension causing spinal cord compression and multiple modalities may have to be used for a single patient.⁹,²⁵ Historically, surgery was the treatment of choice in symptomatic VHs. The aim was spinal cord decompression and sometimes only partial removal of tumor.²⁵ Some surgeons have used laminectomy for spinal cord decompression to treat aggressive VHs.²⁶,²⁷ In some studies, intralesional injection of ethanol has been used successfully. Although there is no long-term follow-up, it were reports of some complications such as pathological fractures, infections, and early recurrences.²⁸ Aich et al.¹⁸ safely performed total excision including a tumor margin in all five patients with aggressive VHs after preoperative transarterial embolization. This treatment allowed good neurological recovery and no recurrence at the long-term follow-up.¹⁰ Balloon kyphoplasty is a developing technique that has successfully been used in the treatment of VHs.¹⁵ Vertebroplasty was first introduced in 1987 by Galimbert and Deramond, French Neurosurgeon and Radiologist respectively, as “alternative” treatment for vertebral hemangiomas.²⁹ Percutaneous vertebroplasty for aggressive and symptomatic VHs even with epidural extension is a valuable, minimally invasive, and quick method that allows a complete and enduring resolution of the painful vertebral symptoms without findings of fracture of a vertebral body adjacent or distant to the one treated.³⁰,³¹ Radiotherapy is used most often to treat lesions associated with local pain,²⁸ but as the sole management modality for patients with progressive neurological deficits it is controversial.² Additional radiation therapy has been recommended after subtotal excision or decompressive surgery.²¹ However, Fox and Onofrio²¹ reported that tumor recurrence was observed in 3 out of 10 patients following subtotal excision and postoperative radiation therapy. Consistent with findings from other reports, the results of our study show that surgery can be performed safely in patients with compressive VHs.¹²,²¹ Posterior decompression with or without instrumented fusion is recommended in patients with total vertebral involvement and circumferential cord compression and when tumor does not involve major vessels or segmental arteries.³²-³⁵ Farrokhi et al.³² reported that inclusion of the fracture level in short segment fixation of thoracolumbar fractures offers better kyphosis correction and fewer instrument failures. Decompression is also recommended in patients with rapid and progressive neurological compromise.¹² An anterior corpectomy and reconstruction is suggested for patients with vertebral body involvement alone and for patients with more extensive vertebral involvement but anterior compression alone.²¹ We prefer a single posterior approach rather than a posterolateral instrumented approach for total excision of spinal tumors in the thoracic spine when the tumor does not involve major vessels or segmental arteries. The reason is that we can observe the spinal cord carefully throughout a single posterior approach, extended to a posterolateral or lateral extracavitary approach. Such
direct view of the spinal cord may not occur satisfactorily during a total en bloc spondylectomy via anterior approach; the complications of which are also more than the posterior approach.34 All patients in this series underwent posterior en bloc resection of the lesion without spondylectomy. This approach offered us with less bleeding and no surgical complications.36 However, some authorities have advocated the use of total en bloc spondylectomy to minimize the risk of tumor spread or residual tumor.37 Postsurgical radiotherapy (RT) is recommended to avoid relapse symptoms that otherwise occur in up to 90% of cases within 3 years.38 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 one month after surgery. There was no recurrence in these patients in 48-month follow up.

In conclusion, laminectomy is a safe and effective procedure for the treatment of cord compression by VH causing stenosis without instability or deformity. Vertebroplasty is useful for improving pain symptoms, especially when vertebral body compression fracture has occurred in patients without neurological deficits. The application of radiotherapy in hemangiomas is to eliminate the abnormal veins and capillaries and to reduce the size of lesion. The results during the long-term follow-up were proved satisfactorily.

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DISCLOSURE

Conflict of interest: None

REFERENCES

Table 1: Patients' characteristics.

<table>
<thead>
<tr>
<th>Patient no.</th>
<th>Age (yrs) and gender</th>
<th>Vertebral body involvement</th>
<th>Chief symptoms</th>
<th>treatment</th>
<th>F/U(6 mo)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>20, F</td>
<td>T4 with posterior element involvement</td>
<td>Weakness (3/5), inability to walk, urinary retention</td>
<td>Posterior laminectomy</td>
<td>Good recovery</td>
</tr>
<tr>
<td>2</td>
<td>48, F</td>
<td>T8 with posterior element involvement</td>
<td>Back pain, sphincter involvement, inability to walk, severe lower extremity weakness</td>
<td>Posterior laminectomy with lateral approach</td>
<td>Walk with help, recovered sphincter</td>
</tr>
<tr>
<td>3</td>
<td>47, M</td>
<td>L1 with pedicle involvement</td>
<td>Severe back pain refractory to medical therapy</td>
<td>Conventional Radiotherapy (3000cGy)</td>
<td>Complete recovery</td>
</tr>
<tr>
<td>4</td>
<td>30, M</td>
<td>Posterior 1/3 body, all posterior element T7</td>
<td>Complete paraplegia, sensory level T7</td>
<td>Laminectomy+ subtotal resection+ radiotherapy+ lateral extracavitary approach</td>
<td>No change</td>
</tr>
<tr>
<td>5</td>
<td>22, M</td>
<td>Body &amp; both pedicles T7</td>
<td>Paraparesis T7</td>
<td>Laminectomy+ irradiation</td>
<td>Walk with help</td>
</tr>
<tr>
<td>6</td>
<td>45, M</td>
<td>Body only L3</td>
<td>Severe low back pain, deformity</td>
<td>Vertebroplasty + spinal fixation</td>
<td>Good recovery</td>
</tr>
<tr>
<td>7</td>
<td>35, F</td>
<td>Body only T9</td>
<td>Mild weakness of lower extremity</td>
<td>Laminectomy with lateral approach+ irradiation</td>
<td>Good recovery</td>
</tr>
<tr>
<td>8</td>
<td>40, F</td>
<td>Body &amp; both pedicles L3</td>
<td>Severe low back pain, Rt lower extremity weakness</td>
<td>Vertebroplasty + spinal fixation</td>
<td>Complete recovery</td>
</tr>
</tbody>
</table>
Table 2: A summary of reported patients of symptomatic vertebral hemangiomas.

<table>
<thead>
<tr>
<th>Author</th>
<th>No. of patients</th>
<th>Age/Gender</th>
<th>Vertebral involvement</th>
<th>Clinical feature</th>
<th>Treatment</th>
<th>F/U</th>
</tr>
</thead>
</table>
| Murugan et al.⁹   | 13              | 15-61y/o   | 12 thoracic, 1 sacral | Backache, myelopathy, radiculopathy, sensory deficits, bladder/bowel involvement | surgery   | 5- become grade I (Ranawat grade)
<pre><code>                                                                    |                                  | 2- one grade improvement       |
</code></pre>
<p>| Kato et al.¹⁰     | 5               | 15, M 51, M 57, F 48, M 64, F | T6 T4/5 T8 T9 T3/4 | Myelopathy, paraparesis, bladder/bowel involvement | En bloc and piecemeal combined total excision | No recurrence, myelopathy improvement |
| Lu et al.¹¹       | 5               | N.A*       | thoracic              | neurological dysfunction | surgery   | No recurrence                           |
| Acosta et al.¹²   | 16              | N.A        | N.A                   | Back pain, neurological deficit | surgical decompression (8) vertebrectomy (8) | Pain relief, improvement in neurological deficit |
| Singh et al.¹³    | 10              | 10-68 y/o  (8 F, 2M) | N.A                   | Paraplegic, sphincter involvement, severe local pain | surgical decompression | bone sclerosis (2) improvement in neurological deficit(10) |
| Urrutia et al.¹⁴  | 4               | 14 M 48 M 72 F 53 M | T12 T6 T5           | paraparesis       | surgery   | complete motor recovery, No recurrence(3) |
| Jones et al.¹⁵    | 2               | 38 M 75 F  | L5 T12               | low back pain, tenderness over the right paraspinal muscles | kyphoplasty | Complete recovery                       |
| Chung et al.¹⁶    | 1               | 47 M       | T6/7                 | back pain, radiating pain to both legs, sensory impairment | total laminotomy | improvement in neurological deficit |
| Alpizar-Aguirre et al.¹⁷ | 1 | neurological manifestation | posterior surgical decompression | good |</p>
<table>
<thead>
<tr>
<th>Study</th>
<th>Cases</th>
<th>Gender</th>
<th>Level of Lesion</th>
<th>Symptoms</th>
<th>Treatment</th>
<th>Outcomes</th>
</tr>
</thead>
<tbody>
<tr>
<td>Evangelopoulos et al.</td>
<td>1</td>
<td>50 M</td>
<td>T8</td>
<td>Back pain</td>
<td>Laminectomy</td>
<td>Improvement of his symptoms</td>
</tr>
<tr>
<td>Pastushyn et al.</td>
<td>64</td>
<td>13 to 78</td>
<td>Cervical, thoracic, lumbar and multilevel</td>
<td>Neurologic signs</td>
<td>Laminectomy with radiotherapy (1000-4000cGy), or vertebrectomy</td>
<td>17-full recovery, 20-useful muscular function, sensory loss, spasticity, abnormal reflex, 10-severe weakness, sensory loss, 13-no recovery, 4-died</td>
</tr>
<tr>
<td>Fox et al.</td>
<td>24</td>
<td>N.A</td>
<td>N.A</td>
<td>Neurological deficit, pain</td>
<td>Decompressive surgery with radiotherapy (1000-4500cGy)</td>
<td>Recurrence in 3</td>
</tr>
</tbody>
</table>

* Not available.