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Management of aggressive vertebral hemangioma with cord compression

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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 or without spinal fixation is suggested for the treatment of 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.[66] 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.

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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, 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

Table 1: The main clinical features and outcome of the study patients

Patient no.	Age in years, gender	Vertebral body involvement	Main clinical features	Treatment	Follow up at 6 months
1	20, F	T4 with posterior element involvement	Weakness (3/5), inability to walk, urinary retention	Posterior laminectomy	Good recovery
2	48, F	T8 with posterior element involvement	Back pain, sphincter involvement, inability to walk, severe lower extremity weakness	Posterior laminectomy with lateral approach	Walk with help, recovered sphincter
3	47, M	L1with pedicle involvement	Severe back pain refractory to medical therapy	Conventional Radiotherapy (3000cGy)	Complete recovery
4	30, M	Posterior 1/3 body, all posterior element T7	Complete paraplegia, sensory level T7	Laminectomy+ subtotal resection+ radiotherapy+ lateral extracavitary approach	No change
5	22, M	Body & both pedicles T7	Paraparesis T7	Laminectomy+ irradiation	Walk with help
6	45, M	Body only L3	Severe low back pain, spinal deformity	Vertebroplasty +spinal fixation	Good recovery
7	35, F	Body only T9	Mild weakness of lower extremity	Laminectomy with lateral approach+ irradiation	Good recovery
8	40, F	Body & both pedicles L3	Severe low back pain, right lower extremity weakness	Vertebroplasty + spinal fixation	Complete recovery

capillary, cavernous or venous blood vessels. Among skeletal locations, vertebrae are second most common site.[7] Incidentally, most VHs, solitary and asymptomatic, are often discovered radiographically. However, local pain, radiological aggressiveness and neurologic deficits may be present.[8] 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).[9-21] 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 Onofrio[21] 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 posteroanterior combined approach for total excision of spinal tumors in the thoracic spine when the tumor does not involve major vessels or segmental arteries.

Table 2: A summary of the patients with symptomatic vertebral hemangiomas reported in the literature

Author	No. of patients	Age in years, gender	Vertebral involvement	Clinical feature	Treatment	Follow up
Murugan *et al.*[9]	13	15-61, 8 M, 5 F	12 thoracic 1 sacral	Backache, myelopathy, radiculopathy, sensory deficits, bladder/bowel involvement	Surgery	5- become grade I (Ranawat grade) 2- one grade improvement
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 improved
Lu *et al.*[11]	5	NA	thoracic	Neurological dysfunction	Surgery	No recurrence
Acosta *et al.*[12]	16	NA	NA	Back pain, neurological deficit	Surgical decompression (8) vertebrectomy (8)	Pain relief, improvement in neurological deficit
Singh *et al.*[13]	10	10-68, 8 F, 2M	NA	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 L5 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
Alpízar-Aguirre *et al.*[17]	1			Neurological manifestation	Posterior surgical decompression	Good
Aich *et al.*[18]	7	39 F 46 F 42 F 33 F 17 F 44 M 31 M	D11 L1 D7-8 D10-11 D12 L1,2-3 D5 D6	Pain, paraplegia, paraparesis	Radiotherapy (4000cGy)	Muscle power improvement

Author	No. of patients	Age in years, gender	Vertebral involvement	Clinical feature	Treatment	Follow up
Evangelopoulos *et al.*[19]	1	50 M	T8	Back pain	Laminectomy	Improvement of symptoms
Pastushyn *et al.*[20]	64	13 to 78	Cervical, thoracic, lumbar and multilevel	Neurologic signs	Laminectomy with radiotherapy (1000-4000cGy), or vertebrectomy	17-full recovery 20-useful muscular function, sensory loss, spasticity, abnormal reflexes, 10-severe weakness, sensory loss 13-no recovery 4-died,
Fox *et al.*[21]	24	NA	NA	Neurological deficit, pain	Decompressive surgery with radiotherapy (1000-4500cGy)	Recurrence in 3

M, male; F, female; NA, Not available.

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] 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. 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. A combination of laminectomy, radiotherapy and vertebroplasty with or without spinal fixation is suggested for the treatment of aggressive VHs with extraosseous extension caused spinal cord compression in all the patients.

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DISCLOSURE

Conflict of interest: None

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