

Aggressive Vertebral Hemangioma Causing Spinal Cord Compression: Presenting a Study of Two Cases and Review of Literature

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Learning Point for this Article:

Aggressive vertebral hemangioma must be considered as one of the differentials in cases of vertebral lesions causing spinal cord compression. Most widely agreed treatment involves pre-operative embolization, followed by surgical decompression, vertebroplasty and post-operative radiotherapy.

Abstract

Introduction: Asymptomatic vertebral hemangiomas are common, but extension into the spinal canal causing cord compression with neurologic symptoms is rare.

Case Reports: Case 1: A 20-year-old male patient presented with difficulty in walking for 6 months with gradually progressive weakness of both the lower limbs. On examination, upper motor neuron signs were present in both the lower limbs with a sensory level below T8 and no bladder involvement. Magnetic resonance imaging (MRI) showed a vascular tumor arising from T6 lamina and pedicle and compressing the cord. Pre-operative computerized tomogram angiography and embolization of the tumor was done, followed by decompression, stabilization of the spine, and vertebroplasty. Postoperatively, the patient received radiotherapy. Case 2: A 71-year-old male patient presented with the recurrence of vertebral hemangioma and cord compression. He had a history of hemangioma with cord compression 13 years back, which was treated by embolization, followed by decompression and fixation. The patient had gradually improved neurologically to normal activities. He was asymptomatic till 7 months back when he noticed difficulty in walking. On examination, pyramidal signs were found to be positive. MRI revealed an expansile lesion at T7 vertebra which was causing compression of the spinal cord. Pre-operative embolization, followed by decompression, stabilization, and vertebroplasty was performed. He also received radiotherapy postoperatively. The diagnosis of benign capillary hemangioma was made after histopathological examination. Neurological recovery was almost complete in both the cases. At 6-month follow-up after surgery, both the patients were able to perform all the activities of daily living.

Conclusion: Aggressive vertebral hemangiomas causing progressive neurological deficit should be treated with surgical decompression, stabilization, and vertebroplasty. Pre-operative angiography, embolization, and post-operative low-dose radiation therapy are recommended.

Keywords: Hemangioma, radiotherapy, thoracic cord compression, vertebroplasty.

Introduction:

Vertebral hemangiomas are benign vascular lesions of bone, composed of multiple thin-walled vessels surrounded by fat infiltrating the medullary cavity between bony trabeculae [1]. Asymptomatic vertebral hemangiomas are common in the spine. However, there are only few case reports of hemangioma growth extending into the spinal canal causing spinal cord compression. Furthermore, the recurrence of hemangioma in

thoracic spine leading to spinal cord compression and myelopathy again at the same site has been rarely reported. With a limited number of cases reported, and characteristic radiological findings not easily identifiable, pre-operative diagnosis is often difficult.

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Author's Photo Gallery



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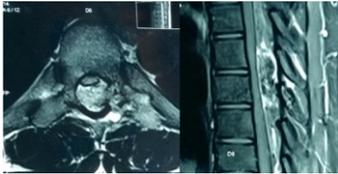


Figure 1: Case 1 pre-operative magnetic resonance imaging (MRI). Axial and sagittal views of MRI showing abnormal tissue growth into the spinal canal, compressing the spinal cord.

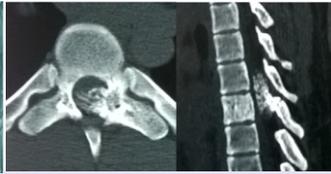


Figure 2: Case 1 pre-operative computerized tomogram (CT) scan. Axial and sagittal views of CT scan showing bony overgrowth from pedicle and posterior elements of T6 vertebra encroaching into the spinal canal, with pathognomonic polka dot pattern in the vertebral body.

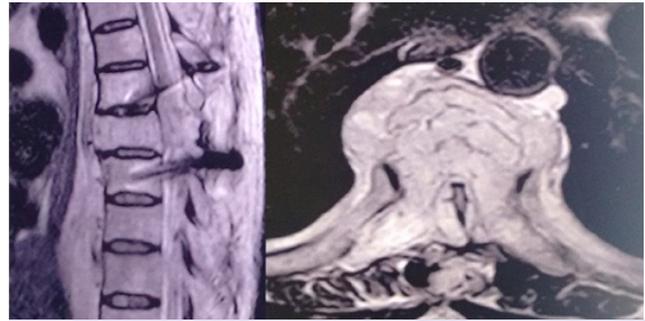


Figure 5: Case 2 pre-operative magnetic resonance imaging (MRI). Axial and sagittal views of MRI showing overgrowth of pathological tissue compressing the spinal cord from all sides.



Figure 3: Case 1 post-operative radiograph. Post-operative radiograph showing decompression and T4 to T8 pedicle screw fixation with vertebroplasty at T6.

Case 1

A male patient aged 20 years presented with insidious onset of difficulty in walking, due to imbalance in gait, for 6 months which was gradually progressive and led to the patient being bedridden for the past 1 month. There was associated back pain, mild dull aching type, and non-radiating, not related with activity, for 6 months. It was also associated with numbness in the lower half of body. Bowel and bladder control was preserved. The patient was admitted to a public hospital before 1 month where he was investigated in the form of magnetic resonance imaging (MRI) and computerized tomogram (CT) scan, which was suggestive of hemangioma. At this stage, the patient presented to us. Nurick Grade 5 and Modified Japanese Orthopaedic Association (JOA) score (using lower extremity score only) was 2. On physical examination, higher motor functions and neurological examination of upper limbs were within the normal limits. Tone of both lower limbs was grossly increased. Power of both lower limbs was Grade 4/5. Touch and pain sensations were impaired

below costal margin. Deep tendon reflexes of both lower limbs were exaggerated with bilateral sustained ankle clonus. Babinski's sign was positive on both sides. Spinal cord compression at T6 was found on MRI (Fig. 1). CT scan revealed heterogeneous

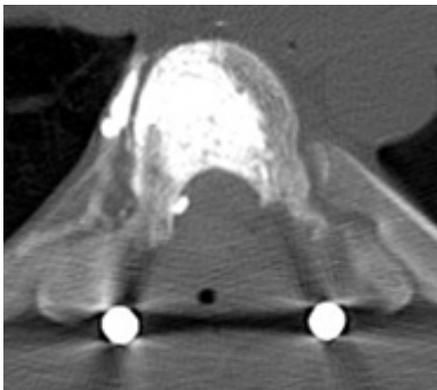


Figure 4: Case 1 post-operative computerized tomogram (CT) scan. Post-operative CT scan showing the spinal canal has been adequately decompressed and vertebroplasty cement filling up the pathological vertebral body.

striated appearance with sclerosis involving T6 vertebral body, as well as its lateral and posterior elements on the left side and bony extension of the same lesion into the spinal canal (Fig. 2). CT angiography and pre-operative embolization of the tumor were carried out, followed by surgical decompression and stabilization. Standard midline posterior approach was used to expose T4 to T8 laminae. T5-6 laminectomy was performed. Bony tumor mass was found arising from left T6 lamina and pedicle; which was carefully drilled out with high-speed burr. Cord decompressed from T5 to T7. Pedicle screw fixation was done from T4 to T8 and vertebroplasty into T6 body (Fig. 3 and 4). Histology of tumor showed benign capillary hemangioma. In the post-operative period, spasticity improved rapidly. The patient was walking with the support of walker from 4th post-operative day. Postoperatively, the patient received low-dose radiotherapy. Neurological recovery was almost complete over a period of 6 months, except for mild numbness over both lower limbs, modified JOA score 7 (using lower extremity score only).

Case 2

A male patient aged 71 years presented with recurrence of hemangioma with cord compression was selected. He had a history of hemangioma with cord compression 13 years back, which was then managed with embolization, followed by decompression and fixation and histologically proved to be benign capillary hemangioma. The patient had gradually improved neurologically to normal activities. He was asymptomatic till 7 months back when he started having difficulty in walking, which was progressive. Bowel and bladder

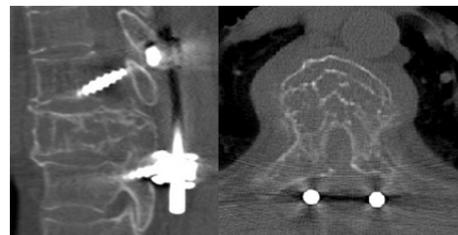


Figure 6: Case 2 computerized tomogram (CT). Axial and sagittal views of CT scan showing expansile lytic lesion in body and pedicles of T7 vertebra, with implants from previous surgery seen in situ.

control was preserved. Nurick Grade 3 and modified JOA score (using lower extremity score only) was 3. On physical examination, the

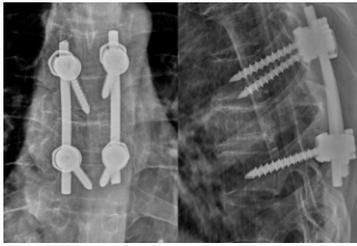


Figure 7: Case 2 Radiograph. Pre-operative radiograph showing implants from previous surgery in situ.

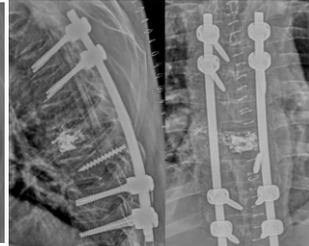


Figure 8: Case 2 post-operative X-ray. Post-operative radiograph showing T4 to T10 pedicle screw fixation with vertebroplasty in affected vertebra T7. Broken part of screw from previous surgery is seen in T8.

patient had myelopathic gait. Higher motor function and upper limb examination were within the normal limits. Both lower limbs were spastic. Motor power Grade was 5/5 in both lower limbs. Hypoesthesia was below T10. Both lower limb deep tendon reflexes were exaggerated; however, Babinski's sign was negative. Radiographic examination showed coarse vertical striations in T7 vertebral body with pedicle screw fixation T6 to T8. MRI and CT scan examination showed bony growth involving T7 body and both pedicles, encasing and compressing the spinal cord (Fig. 5, 6, 7). Embolization was carried out, followed by surgical decompression. With the patient in prone position, midline posterior approach was used to expose T4 to T10 laminae. Pedicles screws and rods of previous surgery were removed. One pedicle screw was broken and had to be left behind. Spinal cord decompressed from T6 to T8 posteriorly and transpedicular decompression at T7. Pedicle screw fixation was done from T4 to T10 and vertebroplasty at T7 (Fig. 8, 9). Histological examination of tumor showed benign capillary hemangioma. Postoperatively, the patient received radiotherapy. Neurological recovery was almost complete (modified JOA score 6) at 6-month follow-up (Fig. 10).

Discussion:

Vertebral hemangiomas have varied presentation, from asymptomatic incidental finding to the rare presentation of spinal cord compression and paraplegia. Asymptomatic

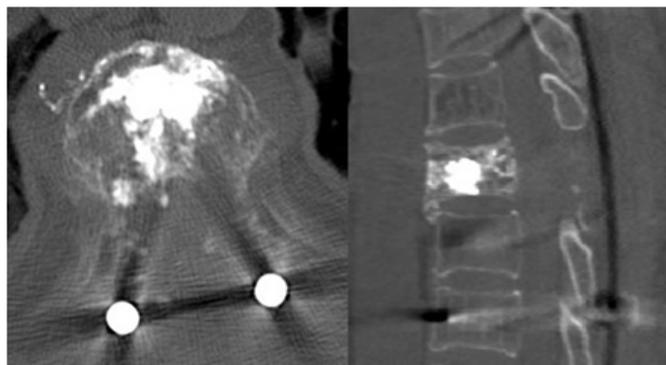


Figure 9: Case 2 computerized tomogram (CT). Post-operative CT scan showing adequate decompression and vertebroplasty cement in situ.

vertebral hemangiomas are common with an estimated incidence of 10–12% of the population based on studies of a large series of autopsies [2]. These are most frequent in the thoracic spine followed by the lumbar spine, although multiple-level involvement is reported to occur in as many as 30% of cases. In contrast, vertebral hemangiomas causing cord compression and neurologic symptoms are extremely rare, and when they occur, are predominantly in the thoracic spine. This locally aggressive subtype of vertebral hemangiomas represents 1–2% of all lesions [3]. Only one level is usually involved [2, 4, 5]. There is no evidence as to why some hemangiomas turn aggressive and become symptomatic while most lesions follow a benign course and remain silent. Symptomatic lesions have been observed to be associated with distinct hyper vascularity [6]; however, biological factors responsible for this have not been understood. Epidemiological studies suggest that patients with symptomatic hemangioma tend to be younger and female, more likely to be found in thoracic spine and in posterior elements. Contrary to this observation, both patients in our study were males, one young and the other being above 70. Pregnancy is considered to be a risk factor for the development of neurological symptoms in patients with previously quiescent hemangiomas. It has been hypothesized that the increase in intra-abdominal pressure caused by growing fetus increases the blood flow to the vertebral venous plexus and that increased estrogen levels may enhance endothelial growth in hemangiomas [2]. Classically, plain radiographs show coarse vertical striations or honeycombing of the vertebral body caused by the presence of thickened trabeculae. This appearance is correlated on CT and MRI as “polka dots.” Computed tomography best defines the bony architecture and is the best diagnostic imaging method [2]. Radiographic appearances of aggressive VHs can be misleading, with features such as extension into the neural arch, irregular trabeculae, lytic areas, poorly defined cortical margins, and soft-tissue extension [5, 6]. In the presence of myelopathy, MRI is the method of choice for imaging the spinal cord and other soft tissue structures. In addition, certain MRI features have prognostic value. Hemangiomas with a high-fat content may represent an inactive

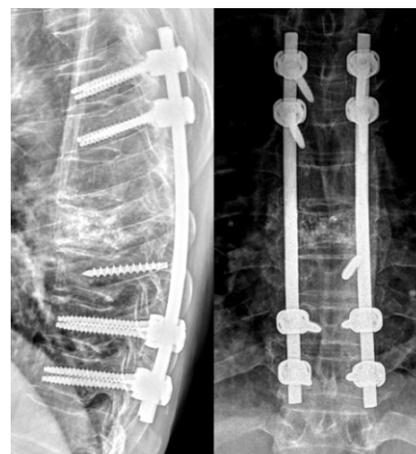


Figure 10: Case 2 follow-up. 2 year follow-up radiograph, showing implants in situ. The patient had a complete neurological recovery at this stage and was completely asymptomatic.

Hemangiomas with a high-fat content may represent an inactive

form of this lesion, whereas lesions with a low-fat content are associated with distinct hyper vascularity and are more likely to cause symptoms of pain or neural compression [6, 7]. Intraosseous signal voids might be an important observation on MRI to suggest vertebral hemangioma [8, 9]. Spinal angiography is both diagnostic and therapeutic in vertebral hemangioma [4, 10]. It helps in determining the vascular supply of both the hemangioma and spinal cord. Endovascular embolization of feeding vessels may be performed before surgery to minimize the risk of massive hemorrhage during surgery. Embolization has been described as sole therapy for compressive vertebral hemangioma [11]. However, few long-term studies on the use of embolization alone have been performed, and other studies have demonstrated the failure of neurologic symptoms to improve after embolization. The progressive neurologic deficit is the main indication for surgical decompression [2, 5]. This ranges from excision of the tumor to laminectomy or corpectomy and toen bloc resection and reconstruction. The choice varies depending on the position and extent of the tumor. Diffuse total vertebral involvement allows only subtotal tumor removal. A common complication of surgical procedures is excessive intraoperative bleeding or post-operative epidural hematoma [2, 12]. This can be reduced by pre-operative transarterial embolization of the lesion [10, 12]. Vertebral hemangiomas are radiosensitive; the lesions have been shown to respond to low-dose radiation. Although radiation therapy is the most common treatment for lesions that cause pain, its use as a sole therapy for patients with progressive neurological deficits is controversial. Most authors have favored surgical decompression, with radiotherapy often used as an adjuvant [2, 10]. Doses of 30–40 Gy are reported to be effective in relieving pain and reducing recurrence after subtotal excision [13, 14]. Although complications including radionecrosis and radiation-induced myelitis have been reported, fractionated doses under 40 Gy spread out over a 3–4 weeks' period are

associated with minimal risk [2]. Sclerotherapy by direct ethanol injection for treatment of compressive vertebral hemangioma has been described. Although some literature suggests good symptomatic relief with sclerotherapy [15], its role is controversial in view of complications including infection, vertebral collapse [15], and Brown-Sequard syndrome [16].

Conclusion:

This case report presents a rare cause of thoracic cord compression. Its clinical manifestations and diagnostic features are outlined. Although rare, vertebral hemangioma with extraosseous extension must be included in the differential diagnosis of cases similar to the current one, particularly when the characteristic trabecular polka-dot pattern is demonstrable, most commonly in the thoracic spine. Computed tomography best defines this diagnostic feature. Surgical decompression with vertebroplasty and post-operative radiotherapy has been successful in patients presenting with progressive neurological deficit. Pre-operative angiography is recommended when the diagnosis of hemangioma has been established and embolization may also be a valuable adjunct.

Clinical Message

Hemangioma must be kept in mind as one of the differentials of thoracic spine lesion with cord compression, because in these cases, it would be imperative to carry out angiography and embolization before undertaking decompression. Vertebroplasty and post-operative low-dose radiation are useful adjuncts for treatment of aggressive vertebral hemangiomas and preventing recurrence.

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