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CASE REPORT

# Thoracic Hemangioma From Rib Presenting as Compressive Paraparesis in a Young Adult

*A Treatment Dilemma*

Ismail Shaik, MS,* Anil Karapurkar, Mch,† Shekhar Bhojraj, MS,* and Premik Naggad, DNB*

**Study Design.** A case report.
**Objective.** To describe the presentation of compressive paraparesis as a result of thoracic rib hemangioma in a young adult and its nonsurgical management.
**Summary of Background Data.** Hemangiomas are rare bone tumors and those arising from rib are rarer. Only about 50 such cases have been reported in literature so far.
**Methods.** A 21-year-old male student, presented to us with a 6-week history of progressive weakness in both lower limbs and loss of bowel bladder control. Patient gave history of being operated for left periscapular tumor treated with wide excision and proven with biopsy to be a hemangioendothelioma (benign but locally aggressive hemangioma variant) a year ago.
**Results.** New radiograph of the chest showed an expansile lesion of left fifth rib and magnetic resonance image showed a tumor of left dorsal thoracic wall with AV malformation causing compressive thoracic myelopathy at T5 level vertebrae. We planned for immediate decompression surgery for spine along with excision of tumor with the help of a thoracic surgeon. However, on preoperative digital subtraction angiography, the tumor was found to be highly vascular with high risk of intraoperative bleeding and morbidity. So, the plan was revised and the patient underwent digital subtraction angiography, followed by embolization by an expert interventional neurosurgeon. The patient showed signs of recovery within a week. Lower limb power improved from grade 2 to 3/5 to grade 4 to 4+/5. The patient became ambulatory with single stick at 3-month follow-up; he was a nonwalker to start with. At 2 years plus follow-up, the patient fully recovered and walks without stick.
**Conclusion.** This unique case brings to light the dilemma a spine surgeon sometimes faces. A case that warranted immediate surgical intervention based on clinical findings was treated with interventional fibrin glue embolizations with excellent results.
**Key words:** arteriovenous malformation, conservative treatment, embolization, hemangioendothelioma., nonsurgical treatment, paraparesis, rib hemangiomas, rib tumors, thoracic hemangiomas, thoracic wall tumors, vascular tumors.
**Level of Evidence:** N/A
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Costal primary tumors are rare and dominated by malignant tumors. Hemangioma of the bone represents only 1% of the bone tumors. Costal localization accounts for only 1% of the cases, and only about 50 cases have been reported in the literature.[1]

Radiological images are not specific and can pose the problem of differential diagnosis with malignant tumors such as osteosarcoma or Ewing sarcoma, benign tumors such as aneurysmal cyst, fibrous dysplasia, or processes such as infectious and parasitic hydatidosis and tuberculosis rib.[1–4] Verification of the blood supply of the spinal cord is highly recommended before resection of a giant tumor from the posterior mediastinum.[5] Chest wall tumors arising near the spinal canal may be associated with enlarged Batson plexus that may hemorrhage during surgical resection.[6]

Available literature mentions very few such chest wall tumors that have caused compressive myelopathy to the spinal cord warranting surgical intervention. Following is an attempt to report one such case of hemangioendothelioma, a benign but locally aggressive hemangioma variant that presented to us as paraparesis secondary to arteriovenous varix compressing thoracic cord.

## CASE REPORT

A 21-year-old male student was admitted with us on August 11, 2012, with weakness in both lower limbs and loss of bowel bladder control for 6 weeks. On neurological examination, power in both lower limbs at hip, knee, and ankle was grade 2/5 (MRC GRADING). He had lost all sensory modalities (pain, touch, proprioception, joint position sense) below T5. His plantars reflex was upgoing (extensor

From the *Wockhardt Hospitals, South Mumbai; Lilavati Hospital and Research Centre, Mumbai; and Breach Candy Hospital, Mumbai, India; and †Breach Candy Hospital, Mumbai, India.

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Address correspondence and reprint requests to Ismail Shaik, MS, Wockhardt Hospitals, Spine Surgery, Nusi Wockhsardt Hospital, Cuncolim, Goa 403703, India; E-mail: ismailwithsmile@gmail.com

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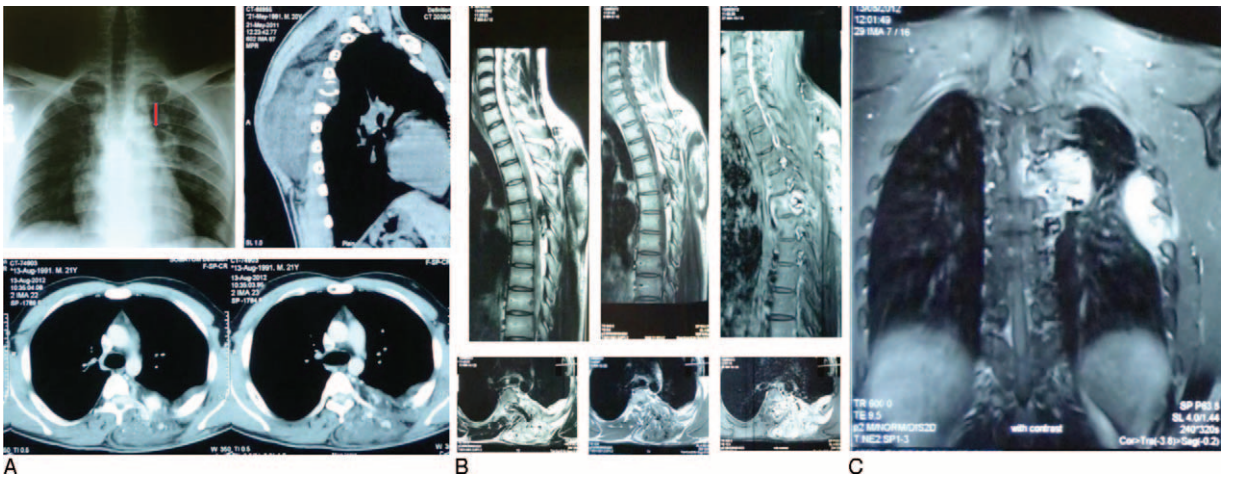


**Figure 1.** HRCT revealed a large vascular malformation with feeding arteries, draining veins on extra pleural space on the left side. Large vascular channels seen in the epidural space compressing the cord. There is expansion and destruction of left fifth and sixth rib on the left. A, Large plaque-like lesion in left hemithorax with destruction of underlying rib and posterior element of D5 vertebra. Extension of the lesion in the left lateral and posterior epidural space causing significant cord compression (B and C).

response) and had brisk knee and ankle reflex with clonus at ankle. According to surgical history, he was operated for left periscapular thoracic wall tumor in July 2011 and proved histopathologically as benign vascular lesion (hemangioendothelioma—a benign but locally aggressive hemangioma variant). New radiograph/computed tomographic scan chest showed a expansile lesion of left fifth rib, and magnetic resonance image showed a tumor of left dorsal thoracic wall with AV malformation causing compressive thoracic myelopathy at T5 level vertebrae (Figure 1). In view of his neurological state, we planned an early decompressive spine surgery, along with thoracic surgeon for thoracic wall tumor excision with prior digital subtraction angiography (DSA) to know the vascular status of tumor and embolize it to reduce intraoperative blood loss. However, on DSA, the tumor turned out highly vascular, with risk of severe and possibly fatal bleeding if operated (Figure 2). It was also concluded that the tumor was paraspinal and not the actual cause of compression. The plan was revised and the patient underwent 3 sittings of DSA-embolizations tissue adhesive glue (embucrylat glue). During embolization, the aim was to deposit the glue within the malformation and occlude only the artery close to the malformation. The proximal artery is kept open as far as possible so as to be able to access it for further embolization if there is a recurrence. The embolizations were each separated by a week on 14th, 21st, and 28th of August 2012. At the end of third embolization, the interventional neurosurgeon was satisfied with the luminal occlusion of feeding artery. The patient also underwent rigorous physiotherapy. The patient improved neurologically (within 3 wk after first embolization). His motor power improved with hip: 4/5, knee: 4/5, and ankle: 4 to 5/5 as per MRC grading. He was able to stand with support. He also recovered a part of his sensory modalities. New magnetic resonance image shows decrease in compression over cord caused by arteriovenous malformation (Figure 3). He went home with a walker support. At 2-year follow-up, the patient has recovered completely and walks independently. His sensory modalities have also recovered and he has regained bowel and bladder control.

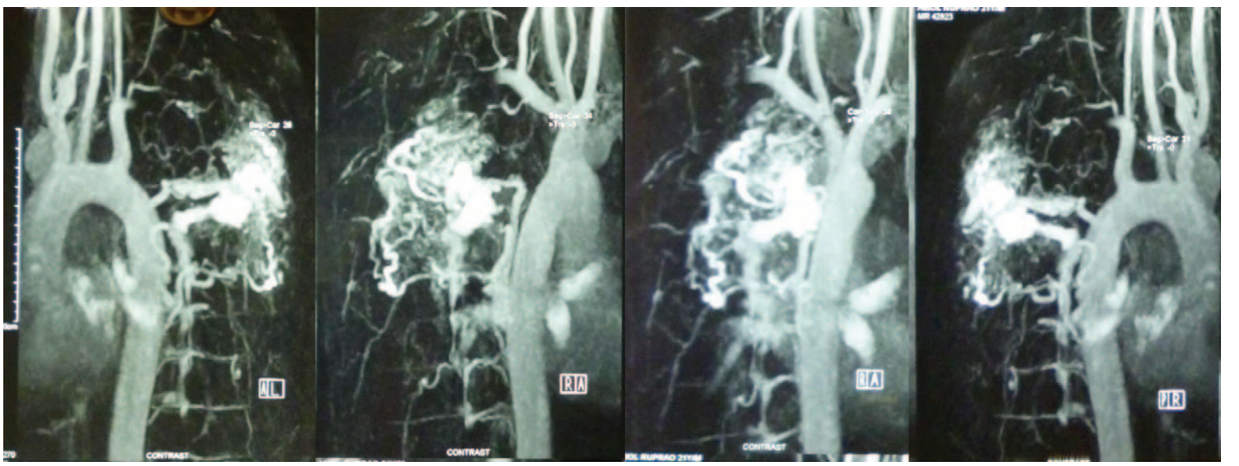

**Figure 2.** The lesion seems highly vascular with vascular supply coming directly from aorta most likely arising from its intercostal branches, as well as large draining veins in the epidural space that drain into the spinal veins.

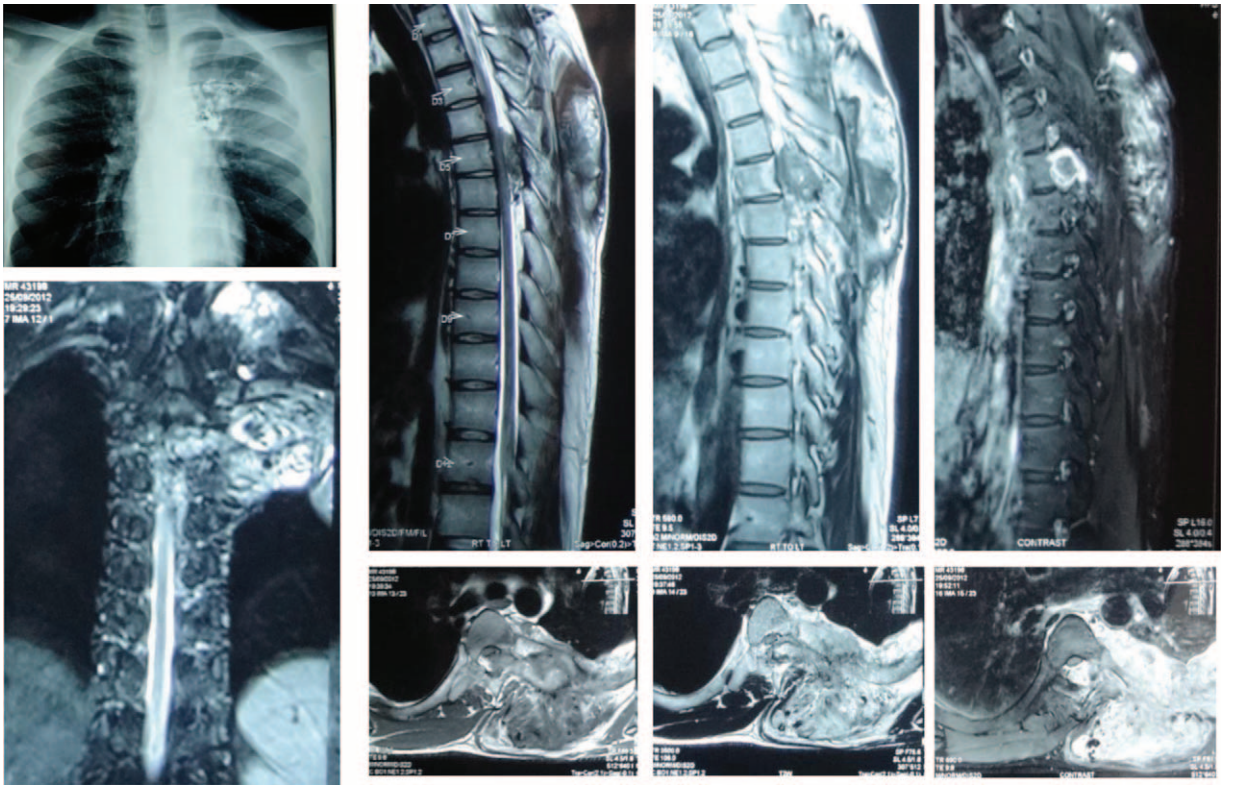

**Figure 3.** Embolic material is seen in the arteriovenous fistula in left upper zone seen on chest radiograph. On contrast magnetic resonance image, there is partial thrombosis of supplying fourth and fifth intercostal arteries and the draining posterior spinal epidural veins of the angiomatous malformation.

## DISCUSSION

This unique case brings to light the dilemma a spine surgeon sometimes faces. To begin with, the patient presented with acute symptoms of cord compression that warranted immediate decompression. However, on digital subtraction angiogram, it became further clear that the tumor is highly vascular with increased risk of bleeding and morbidity if intervened surgically. The actual cause of compression/weakness was the large arteriovenous malformation formed in spinal canal and the pulsatile venous varix that was arterialized. So, the paraparesis was caused partly by mechanical pressure due to malformation and venous congestive edema due to arterialized varix over the cord in the canal. Plan was revised as the paravertebral vascular tumor (hemangioma) was not the cause of paraplegia; hence, excision was ruled out. Also, mechanical pressure was not the only cause for weakness, so decompression would be of limited benefit with risk of uncontrolled bleeding. Hence, spine surgical and cardiothoracic surgical intervention was ruled out. So, the patient was managed with embolizations by an experienced interventional neurosurgeon with excellent results. What was initially planned to be a presurgical workup procedure became definitive treatment modality. The lag of 3 weeks for recovery could be explained as the time needed for the venous congestion around cord to settle after embolization. In conclusion, we would like to propose that if one finds large intraspinal venous varices, one should not operate but do embolizations as an excellent modality of treatment.

### ➢ Key Points

- ❑ Acute paraparesis due to costal/rib hemangiomas close to spinal canal is rare entity.
- ❑ One should always evaluate large intraspinal arteriovenous malformations with digital subtraction angiography to know vascular status of tumor.
- ❑ Embolization is a less invasive way of treating these tumors.

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