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Case Report

LUMBAR VERTEBRAL HEMANGIOMA WITH NEUROLOGICAL INVOLVEMENT: A CASE REPORT AND REVIEW OF THE LITERATURE

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ABSTRACT

A 22 year old girl presented to our outpatient department with weakness and numbness of both lower limbs since last 6 months. On examination lower limb power was MRC 4/5 bilaterally for ankle dorsi flexorss and extensor hallucis longus. She was on provisional ATD since last 4 months. MRI showed hypointense marrow on T1 image and L4 body collapse with posterior elements indenting theca and exiting nerve root. Debridement of body, spinal cord, decompression & stabilization with pedicular screws was carried out through posterior approach. Samples from L_4 body sent for HPE. Section showed spicules of dead and reactive bone surrounding small and large vascular spaces suggestive of vertebral hemangioma. At one year follow up, the patient was pain free with no sensory deficit and improved neurological status to bilateral 5/5 MRC grade for all muscles. Patient was walking without support at that time.

Keywords: *Hemangioma, Neurological Status, Debridement, Pedicle Screw*

INTRODUCTION

A hemangioma is a hamartomatous proliferation of vascular tissue of endothelial origin (Feider and Yuille, 1991). Vertebral hemangioma is the most commonly encountered tumor of the vertebral column (Feider and Yuille, 1991). Cadaveric studies by Topfers and Junghanns (Topfer, 1928; Junghanns, 1932) reported incidence of hemangioma 10-12% in normal population. The age distribution of vertebral hemangioma peaks between 3rd to 4th decades with 2:1 female predominance. Only 0.9-1.2% of all hemangiomas are symptomatic (Healy *et al.*, 1983; Nguyen *et al.*, 1987). Most of them are confined to the thoracic spine and extremely rare in the lumbar area (Pastushyn *et al.*, 1998). Here we present a case of lumbar vertebral hemangioma with collapse of body causing low back pain and neurologic deficit. This article discusses the details of this case, as well as the natural course of vertebral hemangiomas, their management, and literature review of the disease. The patient was informed that the data concerning will be used for publication, and they consented.

CASES

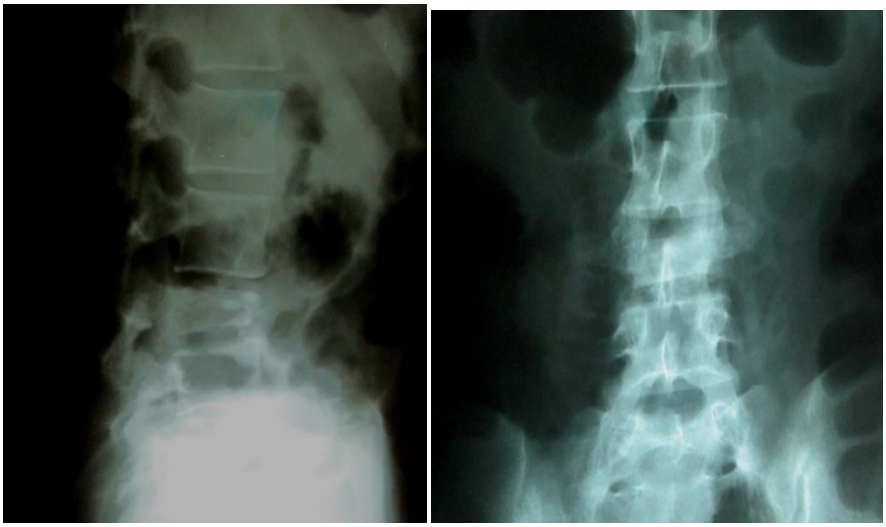
A 22 year old girl presented with severe low back pain with radiation to bilateral lower limb since last 6 months, Progressive numbness of both lower limb since last 6 month and weakness of both the lower limb since last 3 month.

On initial examinations there was tenderness on percussion over lower lumbar spine. Muscle testing showed MRC grading 5/5 for both hip flexors and knee extensors, 4/5 for both ankle dorsiflexor and extensor hallucis longus bilaterally. Diffuse numbness over both lower limb. No blader bowel involvement noted. Patient was on antitubercular therapy since last 4 months.

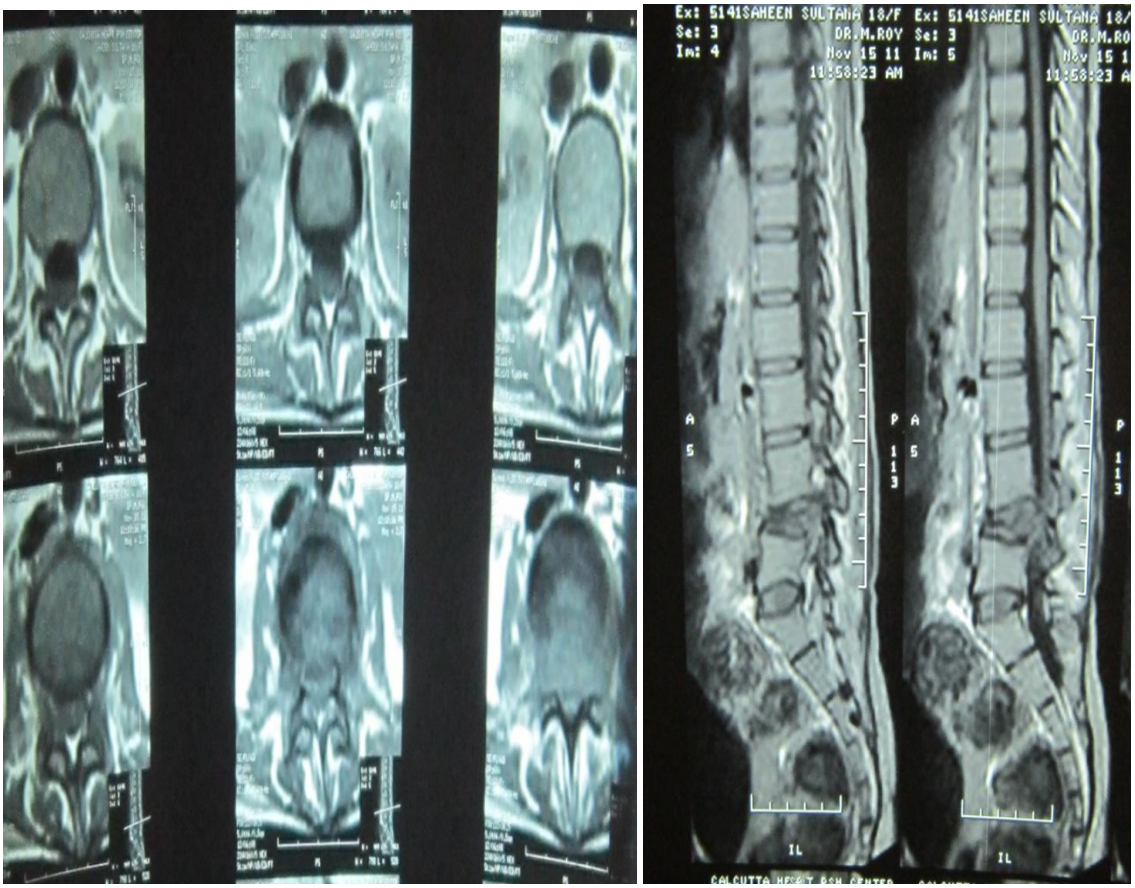
Routine blood investigations were within normal limit. ESR was 20 mm in first hour. CRP 3.3mg/L. Montoux test was positive. Plain x ray [illustration **1, 2**] showed single vertebral (L4) body collapse with normal adjacent disc space. MRI showed hypointense marrow on T1 image [illustration **3, 4**]. L4 body collapse with posterior element indenting theca and exiting nerve root. Intervertebral disc space was normal. Moderate paraspinal fluid collection was seen.

Patient was provisionally diagnosed as Tuberculosis of L4 vertebra and ATD continued. Operative intervention was planned as the neurological signs deteriorated inspite of ATD. Debridement of body,

spinal cord decompression & stabilization with pedicular screws was carried out through posterior approach [illustration **5, 6**].



X ray of lumbar spine AP and Lateral view showing Single vertebra L4 body collapse with Adjacent disc space normal



MRI of lumbar spine showing hypo intense marrow in T1 images and Collapse / compression of L4 body with posterior elements indenting the theca and exiting nerves

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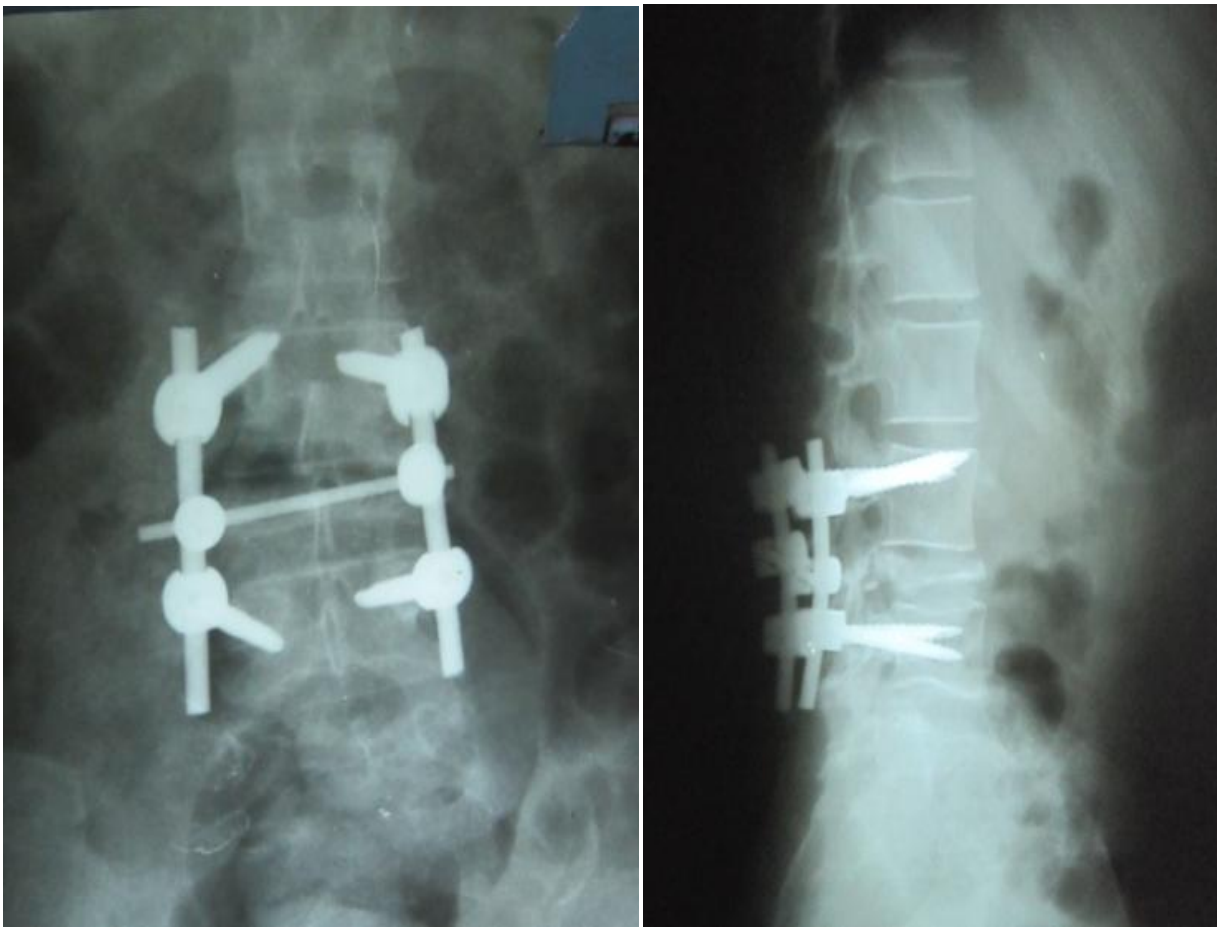
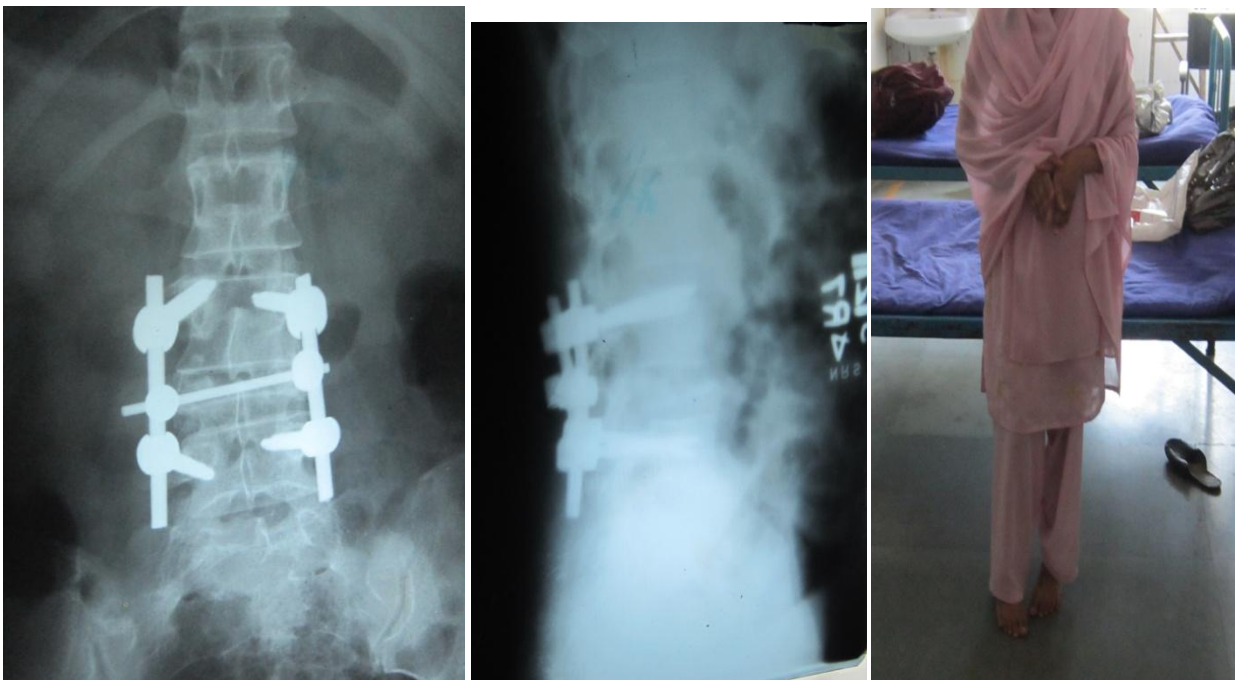


Post operative x ray showing pedicular screws in situ



Post op x-ray and clinical photo after 6 months

Per operatively no pus or casseous material was found. Body was found to be hard. 'Red currant jelly'-like haematoma found in epidural space. Samples from L_4 body sent for HPE. Section showed spicules of dead and reactive bone surrounding small and large vascular spaces, some of which are lined by flattened endothelial cells suggestive of **vertebral heamangioma.**

Patient was referred to dept of radiotherapy no intervention suggested. Followed up at 1 month, 3month and 6 month [illustration **7, 8, 9**] post operatively motor power improved rapidly in both limbs within 1 month and patient was mobilized with braces. At 1 year follow up, the patient was pain free with no sensory deficit and improved neurological status to bilateral 5/5 MRC grade for all muscles. Patient was walking without support at that time.

DISCUSSION

Diagnosing vertebral hemangioma with neurological involvement is very difficult. Clinical presentation and radiology can mimmick anything like meningioma, lymphoma, metastasis, tuberculosis etc.

This is a hamartomatous lesion of vertebral body probably of dysembryogenic origin (Laredo *et al.,* 1986). It can be cavernous, capillary or mixed type. The difference is intervening bone stroma is present in capillary but absent in cavernous type. Pastushyn *et al.,* (1998) reported that 28% of their patients had cavernous type, 50% had capillary type and 22% had mixed type of hemangioma.

Vertebral hemangioma s are classically characterized by sparing and thickening of vertically striated trabecullae which retains the capability to withstand axial load thus called "Jailhouse or corduroy appearance". In axial CT scans, they give the appearance of a "polka dot "or "spikes of bone" pattern because the vertical striations are imaged in cross sections. MRI is now the gold standard of investigations. Quiscent haemangiomas produce high signals on both T1 and T2 images where as haemangiomas demonstrating low T1 and high T2 are active lesions indicating hypervascular lesions with potential to compress the spinal cord. Work ups for aggressive hemangiomas may include angiography to determine vascularity, identify feeding, draining vessels. CT guided biopsy may be warranted to differentiate haemorrhage, lymphoma, myeloma, metastasis etc.

In radiologic evaluation of vertebral hemangiomas, Laredo *et al.,* (1986) described 6 radiographic criteria seen significantly more often in cases of compressive vertebral hemangioma than of asymptomatic vertebral hemangioma. These criteria are thoracic location (T3-9 vertebrae especially); involvement of the entire vertebral body; involvement of the neural arch (particularly pedicles); an irregular, honeycomb appearance; expanded and poorly defined cortex; and swelling of the soft tissue (Laredo *et al.,* 1986).

Symptomatic vertebral haemangiomas are difficult to treat because of the highly vascular nature of the lesion. Four different pathophysiological mechanisms have been documented for symptomatic vertebral haemangiomas (Fox and Onofrio, 1993; McAllister *et al.,* 1975); compression fracture with posterior elements indenting theca was the underlying cause in our case. Distortion of the spinal canal due to tumor enlargement of the vertebral body, tumor extension into the epidural space and bleeding from the mass into the epidural space are the other 3 mechanisms.

There are no randomized control trials, so published case serieses are the only evidences for different treatment options. Management oh vertebral hemangiomas depend on the severity of symptoms (pain and neurological involvements). Painful hemangiomas without neurological involvements can be treated by ballon kyphoplasty, vertebroplasty, selective embolization and radiotherapy but surgical decompression is the most widely accepted mode of treatment for neurological involvement.

Hemangiomas are radiosensitive tumors that responds to 30-40GY and reports have shown symptomatic improvement following radiotherapy (Faria *et al.,* 1985). Miszczyk and Tukıendorf, (2012) reported analgesic effect of radiotherapy for painful vertebral hemangiomas and related this effect to the applied total dose and fraction dose. Faria *et al.,* (1985) reported almost complete disappearance of the symptoms in seventy-seven percent of symptomatic vertebral hemangiomas after radiotherapy. Yang *et al.,* (1985) reported that irradiation could be chosen as the primary treatment of vertebral hemangioma without preceding surgical decompression for patients with a severe compression syndrome of the spinal cord. The beneficial results of radiation therapy have, however, to be weighed against the potential risk of

radionecrosis of the spinal cord and vertebral bodies, leading to a higher incidence of fractures resulting in neurologic deficits in a benign pathology.

Injection of methyl methacrylate (vertebroplasty) is usually performed under fluoroscopic guidance and is indicated for stabilizing vertebral bodies at risk for collapse or to reduce pain. However, cement leakage may occur after vertebroplasty and more complicated surgery may be needed to remove any cement that has leaked (Evangelopoulos *et al.*, 2009). When there is no pathological fracture or the hemangioma is located in the vertebral body not near the spinal canal, the risk of cement leakage is minimal. Vertebroplasty can also be performed preoperatively to make decompression surgery easier because it reduces the risk of hemorrhage (Ide *et al.*, 1996). Balloon kyphoplasty is a developing technique that has successfully been used in the treatment of VHs (Jones *et al.*, 2009).

Endovascular embolization has been used as mainstay therapy for vertebral hemangiomas (Hekster and Endtz, 1987). Trans arterial embolization can also be used preoperatively to minimize bleeding from a vertebral hemangioma during surgery and decrease risk of post operative epidural hematoma (Fox and Onofrio, 1993; Ng *et al.*, 1997). Hekser *et al.*, were first to report reversal of spinal cord compression following percutaneous embolization of feeding vessels (Hekster *et al.*, 1972). Reflus of the embolization material into the lumbar and intercostal arteries may occur and this may lead to spinalcord infarction with paresis (Gross *et al.*, 1976).

Percutaneus alcohol ablation is a newer technique for hemangiomas. Heiss *et al.*, and Bas *et al.*, reported succesful management of spinal cord compression caused by vertebral hemangioma with intralesional alcohol injection (Heiss *et al.*, 1996; Bas *et al.*, 2001). Ethanol reduces the size of hemangioma and allevate cord compression by causing intralesional thrombosis and destruction of endothelium leading to devascularization of the lesion (Doppman *et al.*, 2000). In this technique the needle has to be placed inside vascular space by help of intraoperative angiography. Goyal *et al.*, Munk *et al.*, and Doppman *et al.*, have also reported good results with this procedure (Doppman *et al.*, 2000; Goyal *et al.*, 1999; Munk and Marotta, 1999). Complications like total cord damage, trasient neurological deterioration, pathological fracture, paravertebral abscess and recurrent hemangioma has been reported from their studies. Symptomatic vertebral haemangioma can be treated by vertebroplasty or selective embolization but surgical decompression is the most widely accepted therapy (Pastushyn *et al.*, 1998; Laredo *et al.*, 1986; Fox and Onofrio, 1993; Castel *et al.*, 1999; Galibert *et al.*, 1987; Lee and Hadlow, 1999; Yazici *et al.*, 1996). The aim of surgery is spinal cord decompression and sometimes partial removal of tumor only (McAllister *et al.*, 1975). Posterior decompression with or without instrumented fusion is recommended in patients with total vertebral involvement and circumferential cord compression and when tumor doesn't involve major vessels (Farrokhi *et al.*, 2010; Farrokhi *et al.*, 2012; Kawahara *et al.*, 2009; Farrokhi *et al.*, 2012). In cases of significant vertebral body involvement with epidural estention causing cord compresssion, vertebrectomy or and corpectomy can be done. patients undergoing corpectomy are subjected to instrumentation to provide spinal stability (Fox and Onofrio, 1993; Feurmann *et al.*, 1986; Murugan *et al.*, 2002). Single posterior approach rather than a posteroanterior combined approach for total excision of spinal tumors in the thoracic spine (Ehsanali *et al.*, 2013). because direct view of spinal cord are better In single posterior then total en bloc spondylectomy via anterior approach (Kawahara *et al.*, 2009). The complications of anterior approach also more than posterior approach (Kawahara *et al.*, 2009).

Ours is a retrospectively diagnosed case of vertebral hemangioma where we did laminectomy, debridement and posterior stabilization. We concluded that surgery can be done safely in patients with compressive vertebral hemangiomas.

Consent: Patient has given their informed consent for the case report to be published.

Competing Interest: The author(s) declare that they have no competing interests.

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