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Case report on aggressive hemangioma

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Abstract
Hemangioma is a benign tumor characterized by the proliferation of blood vessels. A few hemangiomas are aggressive, characterized by bone expansion and extra-osseous extension. The vertebral hemangiomas are most often found in the thoracic and lumbar spine. The aggressive type refers to the presence of extension beyond the vertebral body, destruction of the cortex and invasion of the epidural and paravertebral spaces. An aggressive hemangioma can occur at any age with peak prevalence in young adults. We hereby present a case of an Aggressive Hemangioma in a young patient with no co-morbidities exhibiting symptoms of low backache and progressive lower limb weakness and urinary symptoms.

Keywords: Aggressive hemangioma, vertebral hemangioma, venous malformations, angiomatous, hamartomatous, myelopathy, radiculopathy

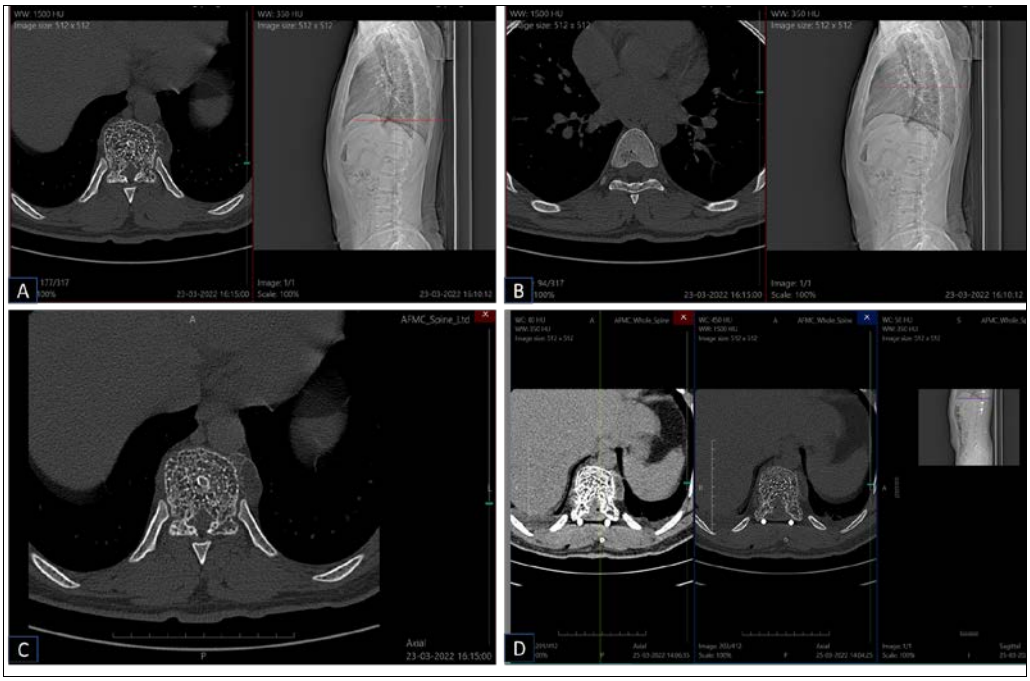
Introduction
Vertebral hemangioma is the most common benign angiomatous lesion involving the spine with an incidence of 10-12%. Hemangioma is a benign tumor characterized by the proliferation of blood vessels. A few hemangiomas are aggressive, characterized by bone expansion and extra-osseous extension. The benign tumors may be mistaken for metastasis resulting in unnecessary biopsies, which have a high risk of hemorrhage. In bone, they are hamartomatous lesions. They have high vascularity, intralesional edema, and intralesional fat. Most hemangiomas are small with about a third being multiple and only 0.9-1.2% of hemangiomas cause symptoms and these are usually aggressive vertebral hemangiomas [4].
The new onset of back pain with subacute progression of thoracic myelopathy has been reported as the most common presentation for patients with a neurological deficit due to aggressive vertebral body hemangiomas. Thus, it is important to recognize this entity and include it in the clinical differential diagnosis and look for the extensions of such vertebral body hemangiomas.

Case presentation
A 24 year old male with no co-morbidities initially presented with complaint of backache of 2 years duration which was insidious in onset, gradually progressive and associated with bilateral lower limb weakness for 5 months. There was also decreased sensation below umbilicus and tightness between lower limbs. History of increased urinary frequency urgency and urge incontinence was also elicited. He was evaluated with MRI dorso-lumbar spine with whole spine screening revealing hemangioma at D10 level with post-contrast enhancement and surgically significant neural compression. Hemangioma was also noted at the D3 level.
On motor examination, bulk was normal, hypertonia in bilateral lower limbs and normal tone in upper limbs were noted. Power was of MRC grade 4 in bilateral lower limbs with ankle clonus. Plantar reflex was bilateral extensor. Sensory examination revealed diminished sensory response to touch, vibration, temperature, and pain below the D10 level with impaired proprioception in bilateral lower limbs. Perianal sensation was diminished with normal anal tone.
After evaluation preoperative embolization was done and pedicle screw fixation of D8, D9, D11, D12 with D10 laminectomy, transpedicular infiltration of polidocanol into the vertebral body of D10 under general anesthesia. A vascular mass lesion involving D10 was noted. 8 screws were placed at D8, D9, D11 and at D12 two rods were placed.

Adequate decompression was achieved with D10 laminectomy. Postoperatively patient was symptomatically better with no fresh neurological deficit. The patient was discharged and kept on follow-up.

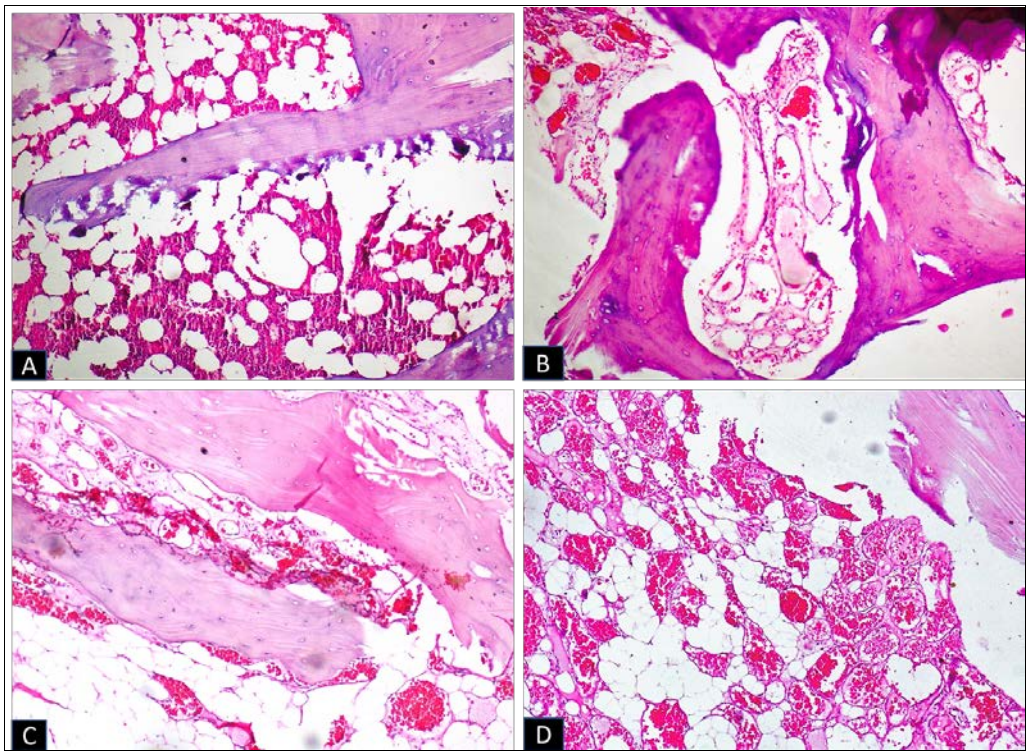
Investigation

MRI LS spine revealed hypodense expansile lesion in D10 vertebra with post-contrast enhancement and surgically significant neural compression with hyperintensity at D10 level suggestive of cord edema. Hypodense expansile lesion at D3 vertebra without significant neural compression. CT scan of spine showed polka dot sign hypodense expansile vertebral body mass in D10 vertebra.

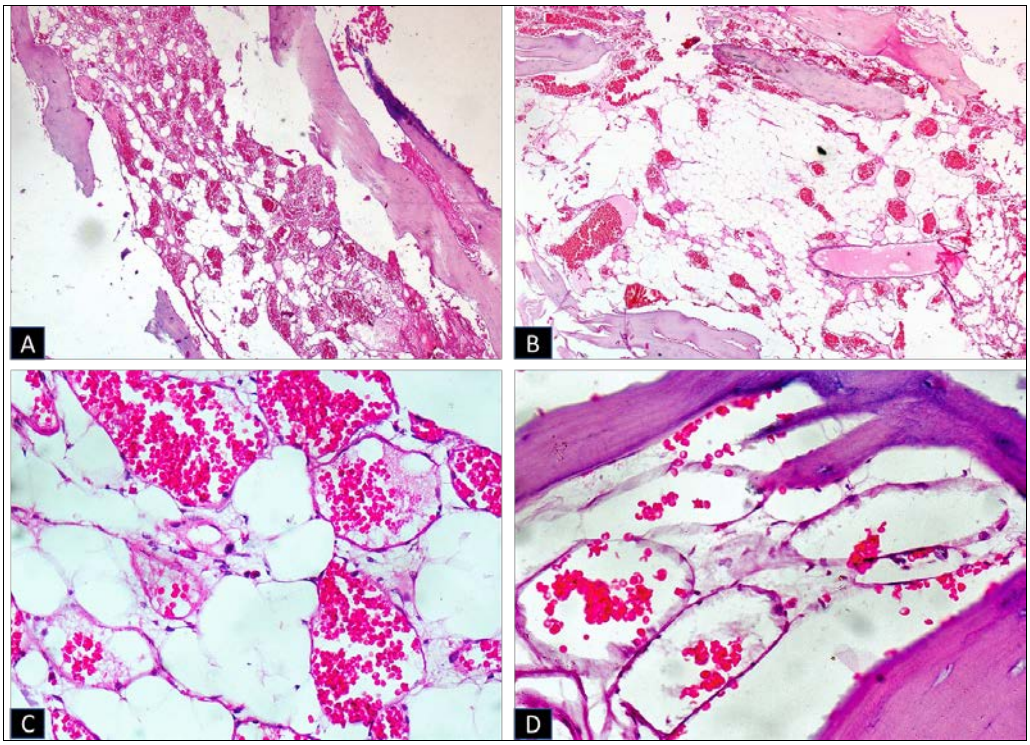


Ct scan imaging – A. Hypodense expansile lesion in D10 vertebra, B. Hypodense expansile lesion in D3 vertebra, C. D10 vertebra showing polka dot sign on CT scan imaging corresponding to vertebral hemangioma, D. Postoperative CT image revealing D10 vascular lesion laminectomy and 8 screws and two rods placement in D8, D9, and D11, D12.

Postoperatively, the laminectomy specimen was sent for histopathological examination. The pathology department received multiple bony tissue bits largest measuring 1.6cm x1.5cm x 0.4cm. Section from the submitted tissue reveals bony trabeculae with numerous thin-walled dilated blood vessels lined by flattened endothelial cells in between bony trabeculae encroaching onto the medullary cavity. Areas of hemorrhage were noted. No atypical or malignant cells were seen. D10 laminectomy specimen was reported as a cavernous hemangioma.



Photomicrograph of laminectomy specimen- bony bits at 100x magnification A. Bony fragment with trabeculae and marrow elements, B. Bony trabeculae with numerous thin-walled dilated blood vessels, C, D. Blood vessels in between bony trabeculae encroaching on to the medullary cavity.



Photomicrograph A. Bony bit at 20x magnification showing numerous thin-walled blood vessels between the trabeculae, B. Dilated blood vessels in the medullary cavity at 40x magnification, C &D. At 100x magnification showing blood vessels lined by flattened endothelial cells with RBCs in the lumen.

Discussion

Hemangiomas are benign vascular neoplasms and can be classified as typical, atypical and aggressive (compressive). The vertebral hemangiomas are most often found in the thoracic and lumbar spine [8]. They are composed of capillary-sized to cavernous blood vessels. The aggressive type refers to the presence of extension beyond the vertebral body, destruction of the cortex and invasion of the epidural and paravertebral spaces. An aggressive hemangioma can occur at any age with peak prevalence in young adults. In the vertebrae, hemangioma is localized preferentially in the thoracic spine.

Neurologic symptoms due to compression of the spinal cord and nerve roots lead to myelopathy or radiculopathy [5, 6, 9]. Clinical worsening and growth during pregnancy is a well-known presentation [7]. The main explanation is vena cava compression and re-routing of blood to the paravertebral, epidural and azygous venous systems.

Histologically, the lesions are composed of fully developed blood vessels with congested dilated vascular channels surrounded by adipose tissue and infiltrating the medullary cavity [2, 9]. The majority of vertebral hemangiomas are historically benign venous malformations characterized by prominent intraosseous abnormal venous channels with flattened endothelium and absence of smooth muscle. The diagnosis of vertebral hemangioma can be made radiologically but aggressive hemangiomas can produce significant osseous expansion and extra-osseous extension such that they can mimic metastasis or hemato-lymphoid neoplasms [9]. The characteristic findings on radiography include coarsened and thickened appearance of vertical bony trabeculae of affected vertebrae. Typical hemangioma on CT imaging show thickened trabeculae and interposed fat yielding a honeycomb appearance on axial images and a striated appearance on sagittal and coronal reconstruction due to the growing soft tissue intermingling with the thick bone trabeculae [4].

Aggressive hemangiomas are hemangiomas with osseous expansion and or extraosseous soft tissue extension contiguous with osseous hemangioma [1, 5, 6, 9]. Differential diagnoses of aggressive hemangiomas include conditions such as malignant and infections. They may become symptomatic. Pain attributed to aggressive hemangiomas may reflect sequelae from osseous expansion or a complicating compression fracture. The extraosseous extension may result in compression of the spinal cord and or nerve roots which may manifest as myelopathy or radiculopathy. When symptomatic management of aggressive hemangiomas frequently requires surgery often with endovascular embolization before surgery to minimize intraoperative blood loss or these lesions can be managed with minimally invasive image-guided techniques including chemical ablation transarterial embolization or radiotherapy [1, 2].

Surgical resection of aggressive hemangioma is usually pursued with large lesions and those with focal neurological deficits [4, 5]. The appropriate surgical procedure depends on multiple factors including lesions location within the spine, location within the affected vertebrae (isolated to the vertebral body, posterior elements, or diffuse), and acuity of symptoms. Endovascular embolization is usually performed to reduce intraoperative blood loss in patients with neurological deficits or those with focal symptoms from their aggressive hemangiomas [4, 5].

Conclusion

Aggressive spinal hemangioma, while histopathologically benign can cause significant morbidity owing to mass effect from marked expansion or extraosseous extension. It is paramount that radiologists recognize the imaging features of aggressive hemangiomas and assess the mass effect on neural elements to expedite appropriate clinical management. Surgery frequently with preoperative embolization is optimal for patients with large lesions and or progressive neurological deficits. Treatment of aggressive hemangioma is a multi-step surgery and requires a multidisciplinary approach [2, 3].

Conflict of Interest

Not available

Financial Support

Not available

Reference

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