Surgical treatment of aggressive vertebral hemangioma causing progressive paraparesis

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2. Case presentation

A 84 years old Italian woman was admitted at our department with a worsening paraparesis started about 6 months before with recent appearance of urinary retention and constipation. Her past medical history was characterized by depressive syndrome, hypercholesterolemia and a surgical procedure for an ovarian benign mass at the age of 50 years. The neurological examination showed a paraparetic gait possible only with external support, proximal muscle strength decrease at D2 and D4 and an old fracture was detected at D7. We performed a surgical procedure of posterior decompression by laminectomy ad D6 with partial extension at D5 and D7; a soft, blu-redish epidural mass was removed and sent for histological examination. During the removal, the lesion appeared highly bloody but we achieved a good hemostasis. No fixation was performed.

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A drainage was positioned and removed at 48h after surgery. No systemic complication, such as deep vein thrombosis, occurred. We started to mobilize the patient on the fourth post-operative day, but it took three days more to achieve the ability to walk with external support for 10–20 m, because of post-operative pain and psychological status. In the following days, her objective neurological status improved gradually, particularly in regard to lower leg muscles strength. However, urinary retention did not improve in the immediate post-operative period. Constipation got progressively better with medial therapy (anthraquinone glycosides). On the third post-surgical day, she performed a control CT scan of the dorsal spine. On the 15th post-operative day, she was transferred to the rehabilitation center. The histological examination showed a vessel proliferation with thin wall and dilated lumen, covered by flat endothelial cells without atypia or mitosis, consistent with cavernous hemangioma. On the basis of histologic examination, clinical status and radiologic imaging, radiotherapy was recommended but the patient did not perform it.
3. Discussion

Despite the benign course of vertebral hemangioma, also known as spinal hemangima or cavernous hemangioma, sometimes his aggressive form may be a challenging lesion to treat and surgery may be required [4,5]. Generally, the lesions, affecting lower thoracic or lumbar spine, are multiple in 30% of cases and affect more frequently post-pubertal females: a hormonal cause has been suggested, thus symptoms often begin during pregnancy, may vary with menstrual cycle and rarely occur before puberty [6]. Exclusive peridural lesion is very rare, but invasion of epidural space after bone involvement can be observed. MRI images help to differentiate typical from aggressive forms before the appearance of symptoms [7]. Vertebral hemangiomas usually show high T1W signal (due to the presence of fat) and high T2W signal (due to the vascular components), but aggressive ones reveal high T2W signal with low T1W signal, cause the reduction of fat content in these cases. Other radiologic findings suggesting aggressiveness are: strong contrast enhancement, cortical erosion, extradural soft tissue, involvement of the posterior elements, presence of stroma in the osseous trabecular and invasion of epidural space. CT scan is the most specific exam for hemangiomas [8] and sagittal images show the typical striated appearance, due to soft tissue growing intermingled with thick bone trabeculae, while axial images show the spotted appearance (the “polka-dot” sign). Trabecular pattern of the involved vertebra has a biomechanic function in preserving the resistance to axial load. Generally, the diagnosis is easily made, but in some cases the hypothesis of malignant lesions, such as vertebral metastasis, myeloma or lymphoma or vertebral infection, should be taken into account [9–12]. Vertebral hemangiomas is generally an asymptomatic incidental finding, but clinical presentation may vary from pain to radicular symptoms and thoracic myelopathy [13–17]. Pain is most often due to the fracture of the involved vertebra or canal stenosis caused by bone expansion, while myelopathy is observed in association with the epidural extension of the lesion. Rarely lesions bleed and symptoms might occur acutely because of epidural hematoma. Management of vertebral hemangioma depends mainly on its clinical presentation [18]. Asymptomatic patients do not require treatment and routine follow-up is not recommended; when diagnosis is uncertain a CT guided biopsy may be performed; this procedure, despite the vascular nature of the lesion, is safe with no bleeding complication reported. Painful lesions refractory to medical therapy, can be managed with radiation therapy [19,20] or, more recently, embolization [21]; both procedures grant a permanent pain relief in 60%–100% of patients, as reported in literature. For patients who present with progressive neurologic deficit, surgical treatment is required. The radical excision is indicated when the lesion involves only posterior elements while, when the vertebral body is involved with bony expansion and prevalent anterior spinal cord compression, anterior corpectomy with graft should be performed. In case of vertebral body involvement with little or no expansion where the spinal cord compression is due to soft tissue invading epidural spaces, the procedure of choice is laminectomy with removal of epidural lesions. Posterior fixation is required when the extent of surgical resection may cause spine instability; however, it is possible that involved vertebral bone is stronger rather than normal vertebra, because of bony expansion and thickened trabecula. In case of multiple contiguous lesions with involvement of anterior and posterior vertebral elements, en bloc total spondilectomy with combined anterior and posterior fixation and fusion has been described [22].

We report a case of thoracic aggressive vertebral hemangioma presenting with thoracic myelopathy. The lesion involved both the vertebral body and the posterior elements with epidural extension. Due to progressive neurologic deficit and the age of patient, we preferred a

![Fig. 3. Post surgery TC scan showing D5 laminectomy. Note the typical trabecula organization of involved vertebra on sagittal images and “polka dot” sign on axial images. Old fracture in vertebra D7.](image-url)
laminctomy with epidural lesion removal without anterior corpectomy also because the spinal cord compression was caused mainly by the epidural mass and not by the bone expansion. In accordance with the principles of biomechanics concerning vertebral fractures [23,24], this clinical case did not require any posterior stabilization system with screws and rods, because the articular processes were spared. Pre-operative embolization may provide reduction of bleeding risks in selected cases [25,26], but in our case embolization was not performed because the specific clinical picture urged rapid treatment and the our surgical approach (laminctomy with lesion removal) exposed to a lower risk of bleeding compared to anterior corpectomy. After the partial lesion removal, radiation therapy was recommended for this patient, because it has been demonstrated to be able to reduce local recurrence, but the patient did not undergo the procedure.

Despite its clinical presentation may be worrying with neurologic deficit, aggressive hemangioma has a satisfactory prognosis if treated with early surgery and, when necessary, radiation therapy and/or embolization. Further studies are needed for the long-term outcome.

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Mauro Dobran: Corresponding author, writer.  
Maurizio Gladi: data collection.  
Fabrizio Mancini: data interpretation.  
Davide Nasi: writer and data collection.  
Stefano Sisti: histological analysis.  
Maurizio Iacoangeli: clinical case evaluation.  
Massimo Scerrati: study concept.

Conflicts of interest  
There are no conflicts of interest to declare.

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Guarantor  
Mauro Dobran.

Consent  
Consent from patient has been obtained.

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