



## Case report

## Surgical treatment of aggressive vertebral hemangioma causing progressive paraparesis

M. Dobran<sup>a,\*</sup>, F. Mancini<sup>a</sup>, D. Nasi<sup>a</sup>, M. Gladi<sup>a</sup>, S. Sisti<sup>b</sup>, M. Scerrati<sup>a</sup><sup>a</sup> Department of Neurosurgery, Università Politecnica delle Marche, Ospedali Riuniti Umberto I, Ancona, Italy<sup>b</sup> Department of Pathological Anatomy, Umberto I General Hospital, Università Politecnica delle Marche, Ancona, Italy

## ARTICLE INFO

## Keywords:

Vertebral hemangioma  
Paraparesis  
Laminectomy  
Cavernous hemangioma

## ABSTRACT

Vertebral hemangioma is a benign vascular lesion that may onset with neurologic symptoms due to spinal cord compression by epidural extension. Surgical procedure, embolization and radiotherapy are the gold standard for the treatment of this disease. We present a case of a 84 years old woman admitted at our department with worsening paraparesis and urinary retention. Her magnetic resonance images (MRI) showed a lesion involving both anterior and posterior vertebral element of D5, with extension into epidural space and spinal cord compression. The patient was operated for laminectomy and epidural lesion removal. Histological examination confirmed the diagnosis of cavernous hemangioma.

## 1. Introduction

Vertebral hemangioma is the most common benign lesion of the spine, most often an asymptomatic incidental finding [1]. Typically, this lesion involves the vertebral body and less commonly it extends to pedicles, lamina or spinous process. Only in 0.9–1.2% of cases it is symptomatic [2] and it is defined as aggressive vertebral hemangioma. In symptomatic cases, generally there is an extension of the lesion into the epidural space with consequent spinal cord compression, but other mechanisms are described such as: spinal canal stenosis, due to the bone expansion, direct compression by vessels feeding the lesion and fracture of the vertebra. Very rarely, deficit may be caused by spinal cord ischemia or hemorrhage with epidural hematoma. Even in cases of aggressive hemangioma, the lesion maintains its benign nature and no malignant degeneration has been reported. The diagnosis is made by computer tomography scan (CT), where we can observe the typical “Polka-dot- sign” due to trabecular aspect of the involved vertebra, and by MRI where the lesion appears hyperintense on both T1WI and T2WI with contrast enhancement. In most cases, vertebral hemangioma has a benign course and surgery is not required. Hereby, we discuss a case of aggressive vertebral hemangioma in a patient suffering with worsening paraparesis that required urgent surgical treatment. The work has been reported in line with the SCARE criteria [3] (see Figs. 1–3).

## 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 deficit at lower leg with (F4-), initial spasticity with positive bilateral Babinski sign, deep tendon reflex exaggerated at the lower leg, tactile hypoesthesia with a sensory level ad D6; the patient also suffered with urinary retention (bladder catheter was positioned) and constipation. The contrast magnetic resonance images (MRI), performed the day after admission, showed homogeneous signal alteration of both vertebral body and posterior arch of D5, with hyper intensity in T2WI; the lesion extended into epidural space causing spinal cord compression and, at the same level, was traced an intramedullary area of T2WI hyperintensity, suggestive for myelomalacia. High, homogeneous contrast enhancing was observed. In addition, other two similar lesions were noted 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-reddish 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.

\* Corresponding author. Clinica di Neurochirurgia, Università Politecnica delle Marche, Ospedali Riuniti di Ancona, Via Conca #71, I – 60020, Torrette di Ancona, Italy.

E-mail addresses: [dobran@libero.it](mailto:dobran@libero.it) (M. Dobran), [Fabrizio.geremia.mancini@gmail.com](mailto:Fabrizio.geremia.mancini@gmail.com) (F. Mancini), [davidenasi83@libero.it](mailto:davidenasi83@libero.it) (D. Nasi), [mauriziogladi@gmail.com](mailto:mauriziogladi@gmail.com) (M. Gladi), [Stefano.Sisti@ospedaliriuniti.marche.it](mailto:Stefano.Sisti@ospedaliriuniti.marche.it) (S. Sisti), [mscerrati@virgilio.it](mailto:mscerrati@virgilio.it) (M. Scerrati).<https://doi.org/10.1016/j.amsu.2017.12.001>

Received 14 August 2017; Received in revised form 22 November 2017; Accepted 4 December 2017

2049-0801/ © 2017 The Author(s). Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).



Fig. 1. Contrast MRI images show signal alteration of the vertebral body with extension to the posterior elements and into epidural space with spinal cord compression.

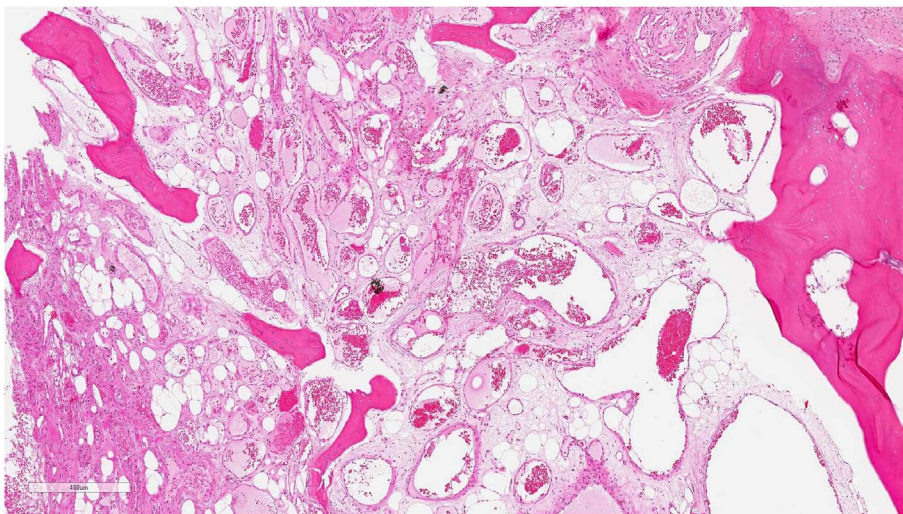


Fig. 2. H&E stain show proliferation of normal vessels with normal endothelial cells, without atypia consistent with cavernous hemangioma.

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.



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.

### 3. Discussion

Despite the benign course of vertebral hemangioma, also known as spinal hemangioma 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



laminectomy 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 (laminectomy 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.

### Ethical approval

No ethical approval was necessary.

### Sources of funding

No source of founding.

### Author contribution

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.

### Registration of research studies

2714.

### Guarantor

Mauro Dobran.

### Consent

Consent from patient has been obtained.

### References

[1] H. Junghanns, G. Schmorl, *The human spine in health and disease*, Grune &

- Stratton, New York, NY, 1971, p. 504.
- [2] J.P. Nguyen, M. Djindjian, A. Gaston, R. Gherardi, N. Benhaïem, J.P. Caron, J. Poirier, Vertebral hemangiomas presenting with neurologic symptoms, *Surg. Neurol.* 27 (4) (1987 Apr) 391–397.
- [3] R.A. Agha, A.J. Fowler, A. Saetta, I. Barai, S. Rajmohan, D.P. Orgill, the SCARE Group, The SCARE Statement: consensus-based surgical case report guidelines, *Int. J. Surg.* 34 (2016) 180–186.
- [4] I.Z. Kosary, J. Braham, I. Shacked, R. Shacked, Spinal epidural hematoma due to hemangioma of vertebra, *Surg. Neurol.* 7 (2) (1977 Feb) 61–62.
- [5] M.W. Fox, B.M. Onofrio, The natural history and management of symptomatic and asymptomatic vertebral hemangiomas, *J. Neurosurg.* 78 (1) (1993 Jan) 36–45.
- [6] I.H. Tekkök, B. Açıköz, S. Sağlam, B. Onol, Vertebral hemangioma symptomatic during pregnancy—report of a case and review of the literature, *Neurosurgery* 32 (2) (1993 Feb) 302–306 discussion 306.
- [7] J.D. Laredo, E. Assouline, F. Gelbert, M. Wybier, J.J. Merland, J.M. Tubiana, Vertebral hemangiomas: fat content as a sign of aggressiveness, *Radiology* 177 (2) (1990 Nov) 467–472.
- [8] J.J. Cross, N.M. Antoun, R.J. Laing, J. Xuereb, Imaging of compressive vertebral haemangiomas, *Eur. Radiol.* 10 (6) (2000) 997–1002.
- [9] S. Leong, H.K. Kok, H. Delaney, J. Feeney, I. Lyburn, P. Munk, W. Torreggiani, The radiologic diagnosis and treatment of typical and atypical bone hemangiomas: current status, *Can. Assoc. Radiol. J.* 67 (1) (2016 Feb) 2–11.
- [10] S.H. McEvoy, M. Farrell, F. Brett, S. Looby, Haemangioma, an uncommon cause of an extradural or intradural extramedullary mass: case series with radiological pathological correlation, *Insights Imaging* 7 (1) (2016 Feb) 87–98.
- [11] M. Dobran, A. Marini, D. Nasi, M. Gladi, V. Liverotti, M.D. Costanza, et al., Risk factors of surgical site infections in instrumented spine surgery, *Surg. Neurol. Int.* 8 (2017 Sep 6) 212, [http://dx.doi.org/10.4103/sni.sni\\_222\\_17](http://dx.doi.org/10.4103/sni.sni_222_17).
- [12] M. Dobran, A. Marini, M. Gladi, D. Nasi, R. Colasanti, R. Benigni, et al., Deep spinal infection in instrumented spinal surgery: diagnostic factors and therapy, *G Chi* 38 (2017) 124–129.
- [13] N.S. Schelezinger, H. Ungar, Hemangioma of vertebra with compression myelopathy, *Am. J. Roentgenol.* 42 (1939) 191–216.
- [14] V.M. McAllister, B.E. Kendall, J.W.D. Bull, Symptomatic vertebral haemangiomas, *Brain* 98 (1) (1975) 71–80.
- [15] R.L. Bell, Hemangioma of a dorsal vertebra with collapse and compression myelopathy, *J. Neurosurg.* 12 (6) (1955) 570–576.
- [16] H.I. Chen, G.G. Heuer, K. Zaghloul, S.L. Simon, J.B. Weigle, M.S. Grady, Lumbar vertebral hemangioma presenting with the acute onset of neurological symptoms, Case report, *J. Neurosurg. Spine* 7 (1) (2007) 80–85.
- [17] H. Ahn, S. Jhaveri, A. Yee, J. Finkelstein, Lumbar vertebral hemangioma causing cauda equina syndrome: a case report, *Spine (Phila Pa)* 30 (21) (2005) E662–E664.
- [18] T.F. Daagi, H.H. Schmidek, Vascular tumors of the spine, in: N. Sundaresan, H.H. Schmidek, A.L. Schiller, et al. (Eds.), *Tumors of the Spine: Diagnosis and Clinical Management*, WB Saunders, Philadelphia, 1990, pp. 181–191.
- [19] S.L. Faria, W.R. Schlupp, H. Chiminazzo Jr., Radiotherapy in the treatment of vertebral hemangiomas, *Int. J. Radiat. Oncol. Biol. Phys.* 11 (2) (1985 Feb) 387–390.
- [20] Pavlovitch JM, Nguyen JP, Djindjian M, Mazon JJ, Piedbois P, Le Bourgeois JP. [Radiotherapy of vertebral hemangioma with neurologic complications].
- [21] L. Picard, S. Bracard, J. Roland, A. Moreno, A. Per, [Embolization of vertebral hemangioma. Technic-indications-results], *Neurochirurgie* 35 (5) (1989) 289–293 305–8.
- [22] R. Ogawa, T. Hikata, S. Mikami, N. Fujita, A. Iwanami, K. Watanabe, K. Ishii, M. Nakamura, Y. Toyama, M. Matsumoto, Total en bloc spondylectomy for locally aggressive vertebral hemangioma causing neurological deficits, *Case Rep. Orthop.* 2015 (2015) 724364.
- [23] M. Dobran, M. Iacoangeli, L.G.M. Di Somma, A. Di Rienzo, R. Colasanti, N. Nocchi, E. Moriconi, D. Nasi, M. Scerrati, Neurological outcome in a series of 58 patients operated for traumatic thoracolumbar spinal cord injuries, *Surg. Neurol. Int.* 5 (Suppl 7) (2014) S329–S332.
- [24] M. Dobran, D. Nasi, D. Brunozzi, L. Di Somma, M. Gladi, M. Iacoangeli, M. Scerrati, Treatment of unstable thoracolumbar junction burst fractures: short-segment pedicle fixation with inclusion of fracture level versus long-term instrumentation, *Acta Neurochir. (Wien)* 158 (10) (2016 Oct) 1883–1889.
- [25] M. Iacoangeli, N. Nocchi, D. Nasi, A. Di Rienzo, M. Dobran, M. Gladi, R. Colasanti, L. Alvaro, G. Polonara, M. Scerrati, Minimally invasive supraorbital key-hole approach for the treatment of anterior cranial fossa meningiomas, *Neurol. Med. Chir. (Tokyo)* 56 (4) (2016) 180–185.
- [26] Y. Robinson, R. Sheta, K. Salci, J. Willander, Blood loss in surgery for aggressive vertebral haemangioma with and without embolisation, *Asian Spine J.* 9 (3) (2015 Jun) 483–491.