Early percutaneous treatment of an aggressive vertebral hemangioma: A case report with a 5-year follow-up

ABSTRACT
Vertebral hemangiomas (VHs) are very common radiological findings, and the majority of them are completely asymptomatic and harmless. However, although rarely, they can present as locally aggressive, symptomatic lesions, and requiring surgery. In these cases, early diagnosis and treatment are mandatory to avoid serious complications and invasive surgery; however, there is no consensus about the best therapeutic option. Minimally-invasive percutaneous techniques have recently gained interests as a therapeutic option. A case of a 58-year-old male with a symptomatic aggressive VH of L5 presenting with untractable low back and radicular pain without neurological deficits is reported. An early percutaneous procedure with selective embolization combined with biportal kyphoplasty of L5 was performed. No complications and a very low-intraoperative bleeding were reported. The patient has been monitored for the following 5 years with a good outcome and with no signs of recurrence. This case report highlights the importance of making the right diagnosis and the advantages of an early percutaneous treatment with selective embolization and augmentation to avoid major open surgery with high risks.

Key words: Balloon kyphoplasty, embolization, percutaneous treatment, vertebral hemangioma

INTRODUCTION
Vertebral hemangiomas (VHs) are very common radiological findings, with a reported prevalence of 10%–12% in the general population. They are considered benign vascular lesions of the bone, often incidentally found at magnetic resonance imaging (MRI), with only the 0.9%–1.2% associated with symptoms such as pain and/or neurological involvement.[1] Hystopathologically, VHs consist in multiple thin-walled vessels, surrounded by fat infiltrating the medullary cavity between bony trabeculae[2] and can be classified as typical, atypical, and aggressive. The term “aggressive” is related to the presence of radiological features such as extension beyond the vertebral body, destruction of the cortex and invasion of the epidural, and paravertebral spaces. Aggressive VHs often require surgical treatment.[3] Currently, evidence suggests the importance of minimally invasive surgeries,[4,5] but no studies reported the combined treatment with balloon kyphoplasty and embolization of the lesion. We report a rare case of aggressive VH of L5 treated with early percutaneous combined techniques at the 5-year follow-up.

CASE REPORT
A 58-year-old male, with a history of chronic low back pain and left sciatica over the past year, was evaluated at our outpatient clinic for the worsening of symptoms.
At the first physical examination, the patient reported severe low back pain and left sciatica irradiated to L5 dermatome (Numeric Rating Scale [NRS]-back = 8, NRS-leg = 7), without neurological deficits.

Standard X-ray [Figure 1], computed tomography (CT) scan [Figure 2], and MRI [Figure 3] of the lumbosacral spine were performed, demonstrating an extensive osteolytic lesion of the soma and left pedicle of L5, involving the posterior wall with intrusion into the spinal canal. Images were suggestive for aggressive VH. Whole-body CT scan, positron-emission tomography-CT, arteriography, and percutaneous vertebral biopsy were performed to exclude a primary bony tumor or metastatic disease. The histological result of the bone biopsy showed a mixed type of VH, characterized by endothelial proliferation, activated endothelial cells, narrow channel in the formation stage [Figure 4] with no evidence of malignancy. The angiographic study [Figure 5] confirmed the presence of a hypervascular lesion of L5 soma with features of aggressive VH mainly fuelled by metameric L5 left artery. The goal of the treatment was to relieve the pain giving a mechanical support to the vertebra. A percutaneous procedure was, therefore, planned to reduce the risks of open surgery. The patient was positioned prone. The neuroradiologist performed a selective embolization of the L5 lesion with the aim of reducing intraoperative bleeding: The common trunk of L5 artery was selectively injected with polyvinyl alcohol particles (contour 150–250 microns), up to the complete disappearance of the pathological vessels [Figure 5]. Subsequently, in local anesthesia, a transpedicular biportal kyphoplasty of L5 was performed. Twenty-mm bilateral balloons were positioned in the L5 vertebral body. The inflation of both balloons increased the available space in the lesion facilitating the following introduction of the cement. Egg-shell technique was used to minimize the risk of cement leakage considering the erosion of the somatic posterior wall: A small amount of polymethylmethacrylate (PMMA) was initially injected;

![Figure 1: Antero-posterior (a) and lateral (b) X-ray of the lumbosacral spine showing an L5 irregular body with structural trabecular alteration](image1.png)

![Figure 2: Lumbosacral computed tomography-scan shows an extensive osteolytic lesion of the body and left pedicle of L5 with involvement of the posterior wall (a) (arrow). The “Polka-dot” signs are suggestive of a vertebral hemangioma (b)](image2.png)

![Figure 3: Lumbosacral magnetic resonance imaging images show a lesion of L5 with hyperintensity in T2 and short-tau inversion recovery-weighted images (a and b), and hypointensity on T1-weighted image (c). Axial short-tau inversion recovery-weighted image (d) show spinal canal invasion and compression of the left L5 root (arrow)](image3.png)

![Figure 4: Histological result (×100) of the vertebral biopsy shows the presence of bony thin layers and clots with optically empty spaces delimited by endothelial lining compatible with mixed vertebral hemangioma](image4.png)
afterward, balloons were slowly reinflated to press the cement against the walls of the cavity, with the aim of repairing the defects in the cortical bone. When the cement eggshell was hardened, the balloons were deflated, removed, and low-pressure cement was inserted using the standard technique. A total of about 7.5 ml PMMA bone cement was used. No intraoperative complications were recorded, and total blood loss was about 50 ml. Postoperative X-rays, CT scan, and MRI were obtained, showing complete filling of the lesion [Figures 6–8]. The low back pain and left sciatica almost disappeared in the early postoperative time, and the patient was discharged from the hospital the day after the procedure. The patient came back to work after 2 weeks. At 5-year follow-up, no radiological or clinical signs of recurrence were reported.

DISCUSSION

No consensus about the best therapeutic option for symptomatic VHs has been found yet. Early differential diagnosis and a correct treatment are the challenges for the surgeons. In case without neurologic deficits, the management includes selective embolization, ethanol sclerotherapy, radiotherapy, and augmentation procedures. In VHs with progressive neurologic deficit, open decompression and fixation followed by local ethanol sclerotherapy are recommended, however, with this technique, serious complications and disease recurrence have been reported. Percutaneous procedures have gained interest in the past decade. There are only few long-term studies about embolization alone in the treatment of aggressive VHs, and they report a high frequency of failures with lesion recurrences requiring open treatment. Recently, interesting results with percutaneous augmentation techniques such as vertebroplasty or kyphoplasty were reported: These procedures provide an immediate mechanical support with durable vertebral stabilization, antalgic effect with quick mobility recovery, and prevented the local recurrences thanks to the local heating generated during PMMA polymerization that kills the remaining tumor cells. Many authors underlined that the balloon kyphoplasty in the treatment of VHs is
associated to less risks of complications as cement leakage than vertebroplasty. In order to reduce the occurrence or recurrences, Hadjiplavou et al. suggested a biportal approach in any case. Preoperative embolization aims at choking off the blood flow in the lesion by blocking the feeding arteries, reducing surgical blood loss, and effectively debulking the lesion due to the induced necrosis. This is the first report at a long-term follow-up of an aggressive VH successfully treated with combined percutaneous techniques. Embolization followed by balloon kyphoplasty could be considered a safe and effective minimally invasive procedure to treat early a symptomatic VH.

Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest
There are no conflicts of interest.

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