CASE REPORT

L1 burst fracture with associated vertebral angioma

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Summary Vertebral angioma is a common bone tumor. We report a case of L1 vertebral angioma revealed by type A3.2 traumatic pathological fracture of the same vertebra. Management comprised emergency percutaneous osteosynthesis and, after stabilization of the multiple trauma, arterial embolization and percutaneous kyphoplasty.
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Introduction

Bone angioma is a benign primary bone tumor characterized by neoformation and agglomeration of abnormal blood vessels.

It is a frequent spinal tumor, affecting 10 to 12% of the population, but is symptomatic in only 0.9 to 1.2% of cases [1,2]; being generally silent, its frequency cannot be determined exactly. Women are two to three times as often affected as men [1,2].

It is a mainly adult tumor, with onset usually between 30 and 60 years of age (range, 2–77 years) [1,2].

The vertebrae and craniofacial bones are involved in some 70% of cases, followed by the femur. In the spine, involvement is principally of the thoracic segment, followed by the lumbar segment. Most reported cases of spinal angioma involved the vertebral body, posterior vertebral arch involvement being more often an extension of an anterior angioma. Several vertebrae may be involved, followed by the ribs and pelvis.

The present study reports a case of pathological vertebral burst fracture with associated vertebral angioma, and discusses treatment options.

Case report

A 39-year-old man sustained multiple trauma in a road accident, with pathologic fracture of L1 without neurologic complications, complex mandibular fracture requiring emergency intermandibular temporary arthrodosis, fracture of both tibial plateaux and thoracic trauma.

CT scan of the dorsolumbar spine found type A3.2 L1 fracture (Fig. 1A–C) with associated angioma of the same vertebra. MRI was not performed, due to the metal used in stabilizing the mandibular fracture.

Given the multiple trauma and the instability of the vertebral fracture, emergency percutaneous osteosynthesis was performed in priority.

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Surgical technique

Step 1
The patient was positioned in ventral decubitus under general anesthesia on a radiotransparent operating table under frontal and lateral fluoroscopy. Percutaneous osteosynthesis used CD Horizon Longitude System uniaxial screws (MEDTRONIC, MN, USA) for type A3.2 L1 fracture reduction by distraction and stem lordosis (Fig. 2A and B). This fracture stabilization allowed surgery for the other traumatic lesions; nursing care was therefore simplified as soon as the assembly was stable.

Step 2
One week later, definitive surgery was performed on the vertebral angioma, with 1-step embolization and kyphoplasty. The patient was positioned in dorsal decubitus, and embolization of both L1 pedicles was performed by the radiologist, using 85 cc ONYX. Control arteriography confirmed complete devascularization of the L1 angioma (Fig. 3A and B). Kyphoplasty was then performed with the patient positioned by the surgeon in ventral decubitus for posterolateral transpedicle percutaneous kyphoplasty of the L1 body (Fig. 4A and B). The bilateral transpedicle approach using 20 mm balloons (Kyphon, MEDTRONIC) allowed a total of 9cc radio-opaque cement to be injected without perivertebral leakage, filling the angioma bone defect.

The patient was raised on the third postoperative day and quickly transferred to a rehabilitation center. At 3 months, he was clinically pain-free. Control CT confirmed good osteosynthesis positioning and leak-free defect filling. Consolidation was achieved (Fig. 5A and B). One-year radiological and clinical examination was satisfactory, with complete return to everyday, occupational and sports activity.

Discussion
Vertebral angioma is a benign lesion formed from normally structured blood vessels, which develop in, deform and
weakens the vertebra or vertebrae, and may secondarily extend into the epidural space.

Being vascular and deforming, vertebral angioma is essentially benign, but may sometimes show aggressive evolution and clinical expression. Prevalence is very high (up to 10% in autopsy series) [3], but clinical expression is rare and neurologic signs even rarer.

Reports in the literature have usually been of small series or case reports. Nguyen et al. found 324 cases in the literature, 139 of which were analyzable and could be compared to a French series of 45 cases [1]. No other large-scale analysis has since been undertaken. The most common location is in the dorsal spine, with a distribution centered around D6 [4]. Multiple vertebral involvement is found in 25% of cases, and is either contiguous or, in 15% of cases, truly multipolar [5]. Neurologic disorder may be induced by several mechanisms, of which the two most frequent are vertebral canal stenosis due to bone deformity in the affected body, or epidural invasion. Fracture of the pathological vertebra or epidural hematoma are rarer [6]. Medullary ischemia induced by vascular steal from the anterior spinal artery has also been reported, but remains anecdotal. Topographic study distinguishes various types of invasion: whole vertebra involvement is the most frequent, but there may also be isolated body or posterior arch involvement or partial invasion straddling the body and the posterior arch [7].

There are various treatment options for aggressive vertebral angioma, but no consensus; strategy is usually a matter of local habit. Total resection is the attitude of choice when feasible [1,8], but is often prevented by technical limitations and/or the extent of invasion.

Whether the surgical approach is anterior or posterior depends on lesion topography. Anterior corpectomy is suited to pure body involvement, where compression is anterior without impact on the posterior spine (24% of cases, according to Djindjian and Nguyen [1,9] the resected body is replaced by an iliac or peroneal bone graft or a prosthesis. In case of posterior arch involvement or associated epiduritis, a posterior approach is used to perform decompression.

Figure 2 Bone-window CT scan, sagittal (A) and coronal reconstruction (B) after percutaneous osteosynthesis: fracture reduction and L1 tumor.

Figure 3 Vertebral arteriography before (A) and after embolization (B).

laminectomy, with greater or lesser articular and pedicular extension [2].

In case of whole body involvement with canal stenosis, attitudes vary, but the best risk-benefit ratio would seem to be associated with extended laminectomy [2].

Extended laminectomy or pediculectomy threatens spinal stability, and posterior arthrodesis has to be associated to the decompression surgery. The main difficulty in vertebral angioma surgery lies in heavy bleeding that may lead to significant blood loss, preventing complete lesion resection [10], especially in corporectomy, where hemostasis is difficult to achieve.

Bleeding, moreover, impairs visualization, with a resultant risk of medullary contusion by instruments in contact with the posterior vertebral wall. Preoperative embolization reduces such risk, without, however, ensuring complete control of hemostasis [11].

Preoperative embolization of the afferent arterial branches feeding the angioma significantly reduces peroperative bleeding; several authors moreover reported neurologic improvement [12,13]. It is thus an especially useful procedure in preparing for surgery. Some authors have recommended it as an isolated treatment for angioma when it resolves the neurological signs; long-term follow-up, however, found recanalization of occluded vessels, with early clinical recurrence [14,15].

The proximity of an artery feeding the medulla or failure to be able to identify the branches feeding the angioma, however, may lead to incomplete embolization and thus to incomplete efficacy [16].

Vertebroplasty by poly-methyl-methacrylate (PMMA) injection relieves spinal pain in 90% of cases [17]. In mixed invasion of the vertebral body and posterior arch, it can achieve consolidation of the body, which is inaccessible to posterior surgery, and reduce bleeding during laminectomy [18]; it may also avoid corporectomy, which is a heavy procedure. It is performed under fluoroscopic or CT control. A large-caliber trocar is introduced on a transpedicular approach, and radio-opaque cement is injected until the angioma is completely occluded, which may require one or several injections. The main complication is a risk of cement leakage into the perivertebral spaces, notably

Figure 4 Bone-window CT scan, sagittal (B) and coronal reconstruction (A): A3.2 L1 fracture.

Figure 5 Postoperative bone-window CT scan, sagittal (B) and coronal reconstruction (A): good PMMA cement positioning without perivertebral leakage.

the radicular foram. In a series of 258 vertebroplasties, including 78 for angiomata, Chiras et al. reported 13 cases of radicular compression by cement leakage [19], with decompression surgery required in three; there was also one case of medullary compression by cement leakage into the spinal canal.

There is also a risk of cement leakage into the epidural veins, which may cause pulmonary embolism; this can be avoided by using a sufficiently viscous cement. Given these possible complications, the procedure should be performed in a surgery center, and is contra-indicated for patients unable to undergo emergency decompression in case of epidural or foraminal leakage [20].

Vertebroplasty is contra-indicated in case of coagulopathy with risk of epidural hematoma. It is also difficult to perform in case of fracture or significant compaction of the vertebral body, and is contra-indicated in case of posterior wall rupture due to the risk of leakage into the vertebral canal. Associated compressive neurologic disorder, although not an absolute contra-indication, calls for the greatest care during cement injection, to avoid any aggravating leakage [20].

Acrylic cement significantly hardens the bone, complicating laminectomy when injected into the posterior arch. In such cases, PMMA injection into the body may be associated to N-butyl-cyanoacrylate or ethanol injection into the posterior arch; both induce in situ angiomata thrombosis, thereby reducing peroperative bleeding without hardening the bone [21].

Ethanol injection was recommended by certain authors as an isolated treatment for angiomata [22–24]. While it achieves destruction of the vascular lesion and regression of neurological signs, however, there was a significant increase in the risk of secondary vertebral impaction [25,26]. Dopman et al., in a series of 11 patients, reported two secondary vertebral fractures requiring surgical stabilization [23], attributed to excessive ethanol quantities. Niemeyer et al. reported one case of Brown-Sequard syndrome secondary to ethanol injection [25], probably due to leakage into the epidural veins, inducing thrombosis. To avoid such complications, Bas et al. injected contrast medium at the outset of the procedure, so as to be able to check that no leakage into the perivertebral spaces was occurring [22].

Acrylic cement can be directly injected through the pedicles during surgery, to consolidate the vertebra [16]. This allows control of any possibly neurotoxic leakage and reduces the risk or per- and postoperative bleeding.

Radiation therapy has also been recommended in aggressive angiomata. Although some authors regard it as an isolated treatment option for vertebral angiomata, its delayed action makes it more suited to cases with mild symptomatology. There have, however, been several reports of neurologic recovery following radiation therapy: Yang et al. reported five cases of recovery, three of which were total, in a series of seven patients with paraplegia of several weeks’ evolution, managed by radiation therapy alone [27]. Despite these reports, radiation therapy is controversial as an isolated treatment in case of significant neurologic impairment; when associated to decompression surgery, on the other hand, radiation therapy shows considerable impact on residual lesions and thus has an essential role to play in the therapeutic arsenal [27]. It also has the advantage of being applicable in elderly and fragile patients. Doses of 35 to 40 Gy spread over 3 to 4 weeks incur a negligible risk of radiculomyelitis [6].

Analysis of the literature enabled us to draw up a treatment strategy for vertebral angiomata inducing neurologic impairment, based on lesion topography. In isolated body involvement (24.4% of cases) [1], surgery may consist in isolated anterior corpectomy, which is, however, a rather heavy procedure, to be reserved to patients with good general health status with signs of severe neurologic impairment by anterior compression of the nerve axis. Otherwise, percutaneous PMMA vertebroplasty to consolidate the vertebral body, followed by radiation therapy in case of active residua, is the attitude of choice. If invasion involves both the vertebral body and the posterior arch (51% of cases) [1], decompression by laminectomy is mandatory; it may be extended to a greater or lesser portion of the posterior arch, and should be associated to osteosynthesis when extensive. Preoperative embolization is useful to reduce bleeding risk; vertebral stabilization can then be achieved by peroperative injection of acrylic cement via a pedicle into the body part of the angiomata. If preoperative embolization is insufficient or unfeasible, percutaneous vertebroplasty by cement injection into the body may be associated to posterior injection of a sclerotic agent to reduce bleeding during laminectomy. In all cases, residual lesions require radiation therapy. If the angiomata principally involves the posterior arch (22.2% of cases) [1], the procedure is basically decompression laminectomy, which may be preceded by embolization or followed by percutaneous injection of a sclerotic agent in case of failure; again, residual lesions threaten neural structures require radiation therapy.

When the neurologic issue has been resolved, there remains that of associated angiomata of varying potential evolutivity. Laredo’s imaging criteria [8] identify such lesions, allowing preventive management.

Radiation therapy appears especially effective in vertebral angiomata, and may be performed as soon as signs of malignancy arise. Where vertebral stability seems threatened by a severe vertebral body lesion, percutaneous vertebroplasty may be associated.

In case of multilevel involvement, preventive measures on several vertebrae may be unduly heavy, notably in terms of radiotoxicity; annual MRI monitoring should then be instituted to detect evolutive lesions before they can induce neurologic damage. This attitude allows targeted preventive treatment of genuinely aggressive lesions.

Conclusion

A search of the literature found no reports of pathologic fracture revealing vertebral angiomata. Minimally invasive management (osteosynthesis, embolization and kyphoplasty) proved able to treat both the traumatic and the tumoral L1 lesion.

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.
References