Case report

Lumbar vertebral haemangioma causing pathological fracture, epidural haemorrhage, and cord compression: a case report and review of literature

S Vinay, SK Khan, JR Braybrooke

Department of Trauma & Orthopaedics, University Hospitals of Leicester NHS Trust, Leicester, UK

Context: Vertebral haemangiomas are recognized to be one of the commonest benign tumours of the vertebral column, occurring mostly in the thoracic spine. The vast majority of these are asymptomatic. Infrequently, these can turn symptomatic and cause neurological deficit (cord compression) through any of four reported mechanisms: (1) epidural extension; (2) expansion of the involved vertebra(e) causing spinal canal stenosis; (3) spontaneous epidural haemorrhage; (4) pathological burst fracture. Thoracic haemangiomas have been reported to be more likely to produce cord compression than lumbar haemangiomas.

Findings: A forty-nine year old male with acute onset spinal cord compression from a pathological fracture in a first lumbar vertebral haemangioma. An MRI delineated the haemangioma and extent of bleeding that caused the cord compression. These were confirmed during surgery and the haematoma was evacuated. The spine was instrumented from T12 to L2, and a cement vertebroplasty was performed intra-operatively. Written consent for publication was obtained from the patient.

Clinical Relevance: The junctional location of the first lumbar vertebra, and the structural weakness from normal bone being replaced by the haemangioma, probably caused it to fracture under axial loading. This pathological fracture caused bleeding from the vascularized bone, resulting in cord compression.

Keywords: Vertebral haemangioma, Pathological fracture, Spinal cord compression, Vertebroplasty, Paralysis, Paraparesis

A 49-year-old, previously fit and healthy man presented to the accident and emergency department, with a history of a few hours of severe lower back pain. This was brought on suddenly after loading a fridge into a van. He had never experienced backache previously, had never been to hospital for any illness in the past, and was not on any medication. The initial examination demonstrated no neurological deficit, with some tenderness over the spinous process of L₁–L₄ with paraspinal muscle spasm. Lumbar spine radiographs (Fig. 1) suggested a wedge-type fracture of the L₁ vertebral body, with 'jailhouse' striations pathognomonic of a vertebral haemangioma. He was admitted for analgesia and neurological observation. He was not commenced on low-molecular-weight heparin, but had leg and foot

pumps applied to both legs as mechanical thromboprophylaxis. His blood work on admission, including a complete blood count, coagulation screen, and electrolytes, were all within normal values for the hospital's laboratory.

Twenty-four hours after admission, a repeat examination showed no neurological deficit, and he was allowed to mobilize as comfort allowed. A CT scan done at this stage confirmed this as an AO type A3.1 (incomplete burst) fracture of L₁, with about 25% loss of vertebral height (Fig. 2). Over the next 24 hours, he developed progressive numbness and weakness in both legs. Examination revealed MRC 1/5 power in left hip flexors, but 0/5 in all other myotomes bilaterally. He had absent knee and ankle reflexes bilaterally, reduced sensation in the right L₃ dermatome, with preserved perianal sensation and anal tone. An urgent MRI of the whole spine demonstrated a haemangioma in the

Correspondence to: SK Khan, 103 Kildale House, Marton Road, Middlesbrough TS4 3TG, UK. Email: Sameer.khan@doctors.net.uk

Figure 1 Lateral radiographs of lumbar spine showing L₁ fracture.

vertebral body, and delineated a large epidural soft tissue component (bleeding from the haemangioma) extending from lower border of T₁₁ to upper border of L₂ (Fig. 3). This was causing deviation and compression of the spinal cord, and resultant significant spinal canal stenosis. No other haemangiomas were seen on these scans.

He was taken to the operating theatre for an emergency spinal cord decompression and stabilization. A large 'red currant jelly'-like haematoma was confirmed in the epidural space, extending from T₁₁ to L₂, and was promptly evacuated. The fracture was stabilized with posterior instrumentation from T₁₂ to L₂ under image intensifier control, with acrylic cement vertebroplasty of L₁ (Fig. 4). There was no extravasation of the cement intra-operatively. A histopathological examination of the biopsy samples from L₁ vertebral body and spinal canal later showed only some degenerative cartilage fragments, but no evidence of malignancy.

His neurological status was closely monitored post-operatively. On the third post-operative day, motor power had improved marginally to MRC grade 1 in bilateral hip flexors and right hip adductor, and grade 2 in left hip adductor group. The rest of his leg muscles were still grade 0, with bilateral knee and ankle areflexia and reduced sensation in L₃-S₁ dermatomal distribution. Over the next 2 weeks, his neurological status improved further to MRC grade 1 in hip flexors/abductors/adductors and knee extensors bilaterally,

Figure 2 CT scan with better definition of the L₁ fracture.

with grade 2 right knee flexion and hallux extension, grade 3 left knee flexion and big toe extension, and grade 3 ankle dorsiflexion and plantar flexion bilaterally. He had a sensory level at T₈ for pinprick, and had no control over his bowel or bladder function.

He was transferred to the regional spinal injuries unit on the 19th post-operative day. He was on bed rest for 6 weeks, and then mobilized on a wheelchair. He learnt to empty his bladder with intermittent catheterization, and was trained to move his bowels over the toilet. Lower limb muscle groups improved to grade 4 on the left, and grade 3 on the right. He was discharged at the end of 3 months with an ankle-foot orthotic. Upon follow-up (6 months post-operative) he had regained reasonable control over his bladder and bowels, lower limb power had improved to grade 4 in all muscle groups except for right ankle dorsiflexors. He was practising walking between parallel bars with the physiotherapists.

Figure 3 Sagittal MRI showing haematoma and extent of bleeding.

Discussion

A haemangioma is a hamartomatous proliferation of vascular tissue of endothelial origin.¹ Its occurrence in the vertebral column was first reported by Virchow.² Reports from cadaveric studies by Topfer³ and Junghanns⁴ described an overall incidence of 10–12%. The lesion most frequently occurs in the vertebral body.⁵ These predominantly affect the thoracic and lumbar vertebrae, with cervical involvement being rarer.^{6,7} Involvement of multiple vertebrae has been reported. Most haemangiomas remain asymptomatic, and do not cause pain or any neurological sequelae. Thoracic vertebral haemangiomas are reported to produce neurological symptoms more often than lumbar. This is thought to be due to the less accommodative ratio of the spinal cord to the canal in the thoracic segment, and the presence of normal thoracic kyphosis, both of which may facilitate early compression.

The neurological manifestations depend upon the cause and degree of root or cord compression, ranging from neuro-tendralgic radiculopathy to complete paralysis. The most frequent mechanisms of cord compression from vertebral haemangiomas are as follows:^{6,8} (1) extension of haemangioma into the epidural space; (2) expansion of involved vertebrae leading to spinal canal narrowing; (3) spontaneous epidural haemorrhage from

Figure 4 Lumbar radiograph showing posterior instrumentation and cement vertebroplasty.

the lesion; and (4) compression fracture of the involved vertebra. Of these, a compression fracture is thought to be rare, because a haemangiomatous vertebra is reinforced by thick sclerotic vertical trabeculae from appositional new bone formation. Certainly in lumbar vertebral haemangiomas, only the first two mechanisms have been documented to cause cord compression.^{9–13}

In this case, the junctional location of the L₁ vertebra, which had already been weakened by the haemangioma, probably led to failure and pathological fracture under axial loading. The patient did not have any abnormal neurological findings at this stage. Mechanical instability then caused bleeding from the highly vascularized bone into the epidural space when the patient was mobilized. This led to cord compression over then next 24 hours. The causation thus combined the last two mechanisms mentioned above.

In 1926, Perman¹⁴ described the radiological features of vertebral haemangiomas, including reduced bone density between much denser vertical trabeculae. This ‘jailhouse’ pattern was reportedly based on the lamellar

arrangement of cancellous bone around space containing vessels. In the thoracolumbar vertebrae the force of gravity in the erect position makes the spongy bone orientate its trabeculae vertically. Numerous radiological studies since then have described typical and atypical features on radiographs and CT scans, corresponding to asymptomatic or locally aggressive haemangiomas.^{15,16} MRI is now the gold standard investigation.^{17,18} Active lesions generate a low T₁ signal and a high T₂ signal, whereas quiescent haemangiomas produce high signals on both T₁ and T₂ images. A vertebral angiogram is recommended if vascularity of the lesion needs to be assessed. Vertebral haemangioma must be distinguished from giant cell tumour of spine, aneurysmal bone cyst and metastatic tumours. It can be used to embolize the lesion, and is discussed below.

The management of vertebral haemangiomas depends on the severity of symptoms (pain, neurological complaints).¹⁹ There are no randomized controlled trials, so evidence for different options comes from published case series. Radiotherapy alone remained the mainstay of treatment for a long time.²⁰ Most studies on radiotherapy alone have incorporated total doses ranging 30–45 Gy, fractionated over a number of weeks. Good to excellent symptom remission rates have been reported.^{21,22} It is debatable whether there is a dose–response relationship²³ or not.²⁴ Radiotherapy after surgical decompression of a symptomatic haemangioma has been noted to be beneficial.²⁵ Repeated irradiation for recurrence may however induce malignant transformation.²⁶

For symptomatic vertebral haemangiomas without neurological compromise, a number of modalities exist nowadays. Transarterial selective embolisation has been studied intensely, focusing on its use as a stand-alone^{27,28} or adjunctive (pre-operative) option. The latter in particular seems to facilitate surgical debulking by reducing haemorrhage and prevents recurrence.^{29–31} An alternative approach is to percutaneously inject ethanol under CT guidance directly into the lesion.^{32,33} While initially considered to be a safe technique, there are reports of infrequent cardiovascular³⁴ and neurological complications.³⁵ Acrylic cement vertebroplasty was first described in 1987,³⁶ and has since been demonstrated to achieve good intraoperative haemostasis and satisfactory stabilization.^{37,38} It has also been used percutaneously before surgery for this purpose.^{39,40} Cement leakage is reportedly rare.⁴¹ Ethanol injection combined with cement vertebroplasty has been successfully used in a series of 12 patients reported by Chen *et al.*⁴²

Surgical intervention is warranted in vertebral haemangiomas presenting with neurological complications.

Decompression of the cord can be accomplished with a laminectomy, or vertebrectomy with instrumentation.^{43,44} Most authors now recommend a systematic approach whereby most symptomatic vertebral haemangiomas are treated with angiography with or without transarterial embolization first, followed by decompression with or without instrumentation, and vertebroplasty.^{45,46}

Conclusions

This pathological fracture occurred in structurally weakened and biomechanically vulnerable vertebra. The fracture should have been investigated with an MRI at presentation to define the pathology and extent of the lesion. Any of the minimally invasive modalities mentioned above (ethanol ablation/embolization/vertebroplasty) would have sufficed at that stage, as there were no neurological sequelae. Vertebral fractures with cord compression warrant urgent surgical decompression, and warrant stabilization with or without vertebroplasty.

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