CLINICAL REPORT

Giant cells tumor recurrence at the third lumbar vertebra

M. Mestiri, M. Bouabdellah, R. Bouzidi, S. Kammoun, N. Mnif, S. Hawet, M. Kooli, M. Zlitni

Department of Orthopaedic Surgery, Charles-Nicole Hospital, Tunis, Tunisia
Department of Radiology, Charles-Nicole Hospital, Tunis, Tunisia
Department of Pathology, La Rabta Hospital, Tunis, Tunisia

Accepted: 17 May 2010

KEYWORDS
Giant cell tumors; Spine; Spondylectomy; Bone tumors prognosis

Summary
Giant cell tumors (GCT) of the spine are rare. We report a case of (GCT) of the third lumbar vertebra revealed by left lumbar radiculopathic thigh pain in a 47 year old man. Imaging showed an osteolytic process invading the vertebral body, the posterior arch and compressing the dural sac left side. Neurological decompression was first performed including stabilization by an instrumented posterolateral graft. A surgical biopsy was obtained at the same time to confirm the diagnosis. A secondary L2-L4 tumor curettage and graft procedure did not prevent, 5 years later, tumor recurrence. We believe that the simple tumor curettage is insufficient to prevent giant cell tumors recurrence.

Introduction
Giant cell tumors (GCT) of the bone are osteolytic lesions, which are usually located in the epiphysis. They are frequent, often recur and benign. Treatment is almost exclusively surgical [1]. Except for the sacrum, spinal forms of GCT are rare [2,3,4,5]. The more severe forms of this entity result in recurrence, malignant degeneration and neurotoxicity [1,2,3,6]. The percentage of local recurrence in the literature is approximately 30% [1]. We present a case of GCT of the third lumbar vertebra initially treated by laminectomy with secondary tumor curettage by the anterior approach. The aim of this study is to present the clinical and radiographic characteristics of these tumors and to discuss therapeutic indications as well as factors favoring recurrence.

Case report
The patient was a 47-year-old man, a nurse, with diabetes and heart disease. He had suddenly developed left L3 lumbar–crural pain 4 months before. Clinical examination showed partial loss of lumbar lordosis and pain during palpation of the L3-L4 vertebrae with paravertebral lumbar...
contraction. The neurological examination showed racket-shaped hypoesthesia of the anterior left thigh, with a minimal motor deficit in the left roots of L3 and L4 which was graded 4 on the motricity test. Genito-sphincter results were normal. The rest of the clinical tests and the biological results were normal. Because a rapid magnetic resonance imaging examination (MRI) was impossible, a CT myelogram was performed. A standard anterior-posterior X-ray (Fig. 1a) and lateral view (Fig. 1b) associated with a myelogram showed purely osteolytic bone deficit with unclear boundaries associated with collapse of the posterolateral third lumbar vertebra and erosion of the left vertebral pedicle resulting in a "one-eyed" vertebra. This was associated with incomplete extradural uptake in the spine, which was opaque (across from L3).

Axial CT myelogram (Fig. 2a) with sagittal reconstruction (Fig. 2b) confirmed the presence of well-limited osteolytic lesions of dense tissue of the hemibody of L3 associated with left anterolateral epiduritis compressing and forcing out the dural sheath. The patient first underwent decompression laminectomy and L1-L4 osteosynthesis as well as a transpedicular biopsy of the tumor. The post-operative outcome was uneventful and the spine was immobilized in a brace. Pathological analysis of the biopsy sample showed rich collagen with numerous cells and fibroblasts, mononucleate cells and giant cells. There were no cytological signs of malignancy. A diagnosis of grade 1 Sanerkin’s GCT was made [7].

Two months after surgery lumbar pain had disappeared and crural pain had markedly improved. MRI (Fig. 3a,b) results confirmed the CT Scan results and showed a high intensity signal on T2 and low intensity on T1 sequences with moderate, heterogeneous enhancement of the left hemibody of the L3 vertebra. Invasion of the anterior epidural space and compression of the dural sheath was also seen. A second procedure by anterior approach included emptying the tumor by curettage and suction and discectomy of L2-L3, L3-L4. Macroscopically excision of the tumor was

**Figure 1**  a: osteolytic bone deficit with unclear boundaries and vertebral collapse, incomplete extradural uptake on the opaque spine; b: invasion of the left pedicle of the vertebra (one-eyed vertebra).

**Figure 2**  a: axial CT Scan: localized osteolysis of the body and posterior arch with invasion of the canal; b: sagittal CT Scan: compression of the dural sheath.
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Figure 3  a,b: MRI: high intensity signal of the lesion on T2–weighted sequence (a), low intensity signal on T1 sequence (b) and moderate heterogeneous enhancement of the hemibody of the left L3 vertebra, invasion of the anterior epidural space and compression of the dural sheath.

considered complete. Marked peridural bleeding was controlled with careful hemostasis. The interbody bone graft was performed with two free fibular transfers. The postoperative course was uneventful, and lumbar-crural pain completely disappeared. The patient was followed-up every three months the first year, every six months the second and third years and yearly thereafter. Clinical and radiographic controls were performed during follow-up visits. At 5 years of follow-up, the patient consulted for progressively developing weakness in both legs, which limited walking to 500 meters. The neurological examination showed grade four motor paraparesis. Standard anteroposterior (Fig. 4a) and lateral (Fig. 4b) X-rays showed osteolysis around the fibular graft with segmental lumbar kyphosis and collapse of L5. Results of MRI strongly suggested tumoral recurrence (Fig. 5a-b). CT scan guided puncture biopsy confirmed the presence of Sanerkin’s grade 1 GCT [7].

Discussion

Spinal GCTs are rare; according to the Mayo clinic, they represent 6.5% of bone GCTs [6] and 1—9% of bone GCTs according Bedwell et al. [8]. The largest worldwide series of 31 cases of giant cell tumors from different centers was reported in 1977 by Dahlin et al. [2]. In 1993, Sanjay et al. [6] reported 24 cases of spinal giant cell tumors from cases in the Mayo Clinic between 1955 and 1989. Although this lesion often only involves one vertebral, Kos et al. [9] published a case of multifocal thoracic and sacral spine GCT and Erdogan et al. [4] published a case of GCT in the sixth cervical vertebra. The different reported cases often occurred in patients between 20 and 30 years old and especially in women [2,6,10,11].

Spinal pain with or without radiculalgia is the most frequent cause for consulting [3,6,10]. However, the diagnosis of GCT is often made after a neurological deficit has developed [2,3,6,10]. Standard X-ray usually shows an osteolytic lesion, without peripheral bone condensation, of even density, which follows the intervertebral disc. If no pedicle is identified on anteroposterior X-ray views ("one-eyed" vertebra), this suggests invasion of the posterior arch. Unlike an aneurysmal bone cyst, GCT usually develops in the vertebral body then invades the posterior arch [10]. In the case reported here, a CT myelogram was performed because a rapid MRI was impossible. CT scan is useful to evaluate bone degeneration because of the excellent spatial resolution. A precise evaluation of any epidural invasion associated

Figure 4  a,b: Standard X-ray: osteolysis around the fibular graft with segmental lumbar kyphosis and collapse of L5.
Histological confirmation of the diagnosis requires a surgical biopsy [2] or a CT scan guided puncture biopsy, whose reliability is 65% [15]. In this report we performed a surgical biopsy at the same time as decompressive laminectomy. A CT scan guided puncture biopsy is safe [16], and was performed to confirm tumor recurrence. In most cases, the histological examination confirms the diagnosis of GCT and excludes the main differential diagnoses, in particular aneurysmal cyst [1]. Sanerkin is the reference classification for the histological grading of a bone GCT [1,17]. Grade I is the benign form of the disease, while grade III is osteosarcoma, and grade II is a borderline form.

Treatment of these tumors must take into account three problems: mechanical because of the extensive osteolysis of the vertebral body, neurological and tumoral with the risk of recurrence [2,6,11]. Treatment of spinal GCT is usually surgical [1,2,3,5,6,10,11]. The possibilities of extratumoral surgery are extremely limited [18]. An isolated lesion of the vertebral body can be treated by total spondylectomy by the anterolateral approach [18,19,20]. Unfortunately, extension into one of the two pedicles makes extratumoral resection impossible [18]. Partial spondylectomy, corporectomy or resection of the posterior arch is a viable option in well-circumscribed lesions [2,6,11,18,19,20,21,22,23]. In a GCT of the second lumbar vertebra which had invaded the medullary canal, Li et al. [3] performed wide en bloc resection of the tumor, including the vertebral body and the psoas muscle followed by a laminarthrectomy by resection of the right root of L2. No recurrence had occurred at 13 years of follow-up. Several vertebral reconstruction procedures have been used. Lafarge et al. [2] filled bone defects with autologous grafts alternating with slices of allograft strengthened with transversal screws and screw plate osteosynthesis. Li et al. [3] used fibular grafts to strengthen vertebrae above and below with compression screws. Smarts et al. [19] performed posterior resection and short-term osteosynthesis, then anterior corporectomy with a cage implant for filling, then a posterior approach for pedicular reconstruction. The use of adjuvant radiotherapy is considered to be a factor favoring the development of sarcoma in an estimated 10% of cases [6]. It can be indicated in inoperable GCT [10,24], incomplete GCT resections, recurrent GCT [6] or as adjuvant therapy to surgery [21,24]. The role of biphosphonates in the prevention of recurrent bone GCT was confirmed in a study by Tse et al. [25]. Its efficacy in spinal forms was reported by Fujimoto et al. [26] but in association with radiotherapy.

Bleeding during surgery of spinal GCT is a severe complication, which can make it impossible to complete the surgical procedure [11]. Preoperative embolisation can prevent this complication and reduce the size of the tumor, facilitating resection [27].

Recurrent GCT after surgery is a serious complication, and treatment is a problem. Most authors believe that it is due to marginal surgical resection [3,28]. Sanjay et al. [6] reported 10 cases of recurrence in 24 spinal GCT. According to Campanacci et al. [29], 90% of recurrence developed in the first three years after surgery. He noted that recurrence had not occurred in total spondylectomy 13 years after surgery. In our report, recurrence developed 5 years after surgery at a stage of neurological compression. Follow-up visits ought to have included MRI imaging to detect recurrence as early as possible. The complication in our report is mainly explained by insufficient resection, which was limited to simple anterior curettage. Recently, Junming et al. [21] published a series of 22 cervical spine GCTs. The rate of recurrence with subtotal spondylectomy was 71% while for total spondylectomy it was only 7.7%.
Conclusion

GCTs of the lumbar spine are rare and their clinical and radiographic characteristics are not specific. MRI is indispensable to evaluate local extension and especially to identify nerve compression. If the vertebral body and the posterior arch are affected, curettage of the lesion is insufficient to prevent tumor recurrence. This occurred in the present report, where a total spondylectomy should have been performed to minimize this risk.

Conflict of interest statement

There is no conflict of interest.

References