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Case Report

Chondroblastoma of the Lumbar Vertebra Associated with Cauda Equina Compression
腰椎的軟骨母細胞瘤併發馬尾狀神經群受壓——病例報告

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ABSTRACT

Chondroblastoma is a benign tumour, most often affecting the epiphyses of long tubular bones such as the proximal end of the humerus, femur, and tibia, as well as the distal end of the femur. Vertebral involvement is extremely rare. We report a case of chondroblastoma of the second lumbar vertebra associated with cauda equina compression. Complete excision is necessary to relieve the compression and ensure surgical clearance.

中 文 摘 要

軟骨母細胞瘤是一種良性腫瘤，最常發病於長管狀骨的骨骺處，如肱骨、股骨和脛骨近端並股骨遠端。椎體發病是極為罕見的。我們報告一病例，其軟骨母細胞瘤在第二腰椎(L2) 發病併發馬尾狀神經群受壓。完全切除術是作為解壓和外科清除的必需手段。

Introduction

Chondroblastoma is a benign tumour, arising most often in the epiphyses of long tubular bones. It was first described by Ewing in 1928 as a calcifying giant cell tumour (GCT) and found by Codman in 1931 to be of cartilaginous origin. It is uncommon, accounting for about 1% of primary benign bone tumours. It is derived from cartilage germ cells or cells of the epiphyseal cartilage. The common locations of these tumours, in descending order, are the proximal end of the humerus, the distal end of the femur, the proximal end of the femur, the proximal end of the tibia, the talus, and the innominate bone.[1,2] It is considered benign with a good prognosis in the majority of cases but metastatic spread has been reported.[3] Vertebral involvement is extremely rare, with fewer than 10 cases reported in the literature.[4] There is a slight male preponderance. It may occur at any age but the majority are in the 2nd decade of life.[1,2,5] Pain is the most common presentation symptom.[6]

Case Report

A 27-year-old Chinese lady presented with a 1-month history of low back pain. She also complained of weakness, pain, and numbness of both lower limbs for 1 week prior to the consultation. The right leg was affected more than the left leg, and the symptoms were progressing. The pain was not related to activity or posture. She denied any history of precipitating trauma. There was no loss of appetite, change of body weight, fever, or night pain. Bowel and bladder control were satisfactory. Her past medical health was good.

On physical examination, she was slim and well nourished. There was loss of lumbar lordosis but no tenderness on her spine. The lower limbs power was Grade 4 at her hips, Grade 3 at her knees and ankles. She was in a wheelchair. There was no sensory loss and the lower limb reflexes were normal with the Babinski reflex downgoing. *Per rectum* examination was normal.

The laboratory investigations were unremarkable with an erythrocyte sedimentation rate of 10 mm/hour. X-rays showed an expansile lesion in the second lumbar vertebra (L2) and loss of lumbar lordosis (Figure 1A and B). A computed tomography (CT) scan showed a lytic lesion in the L2 vertebra with nerve

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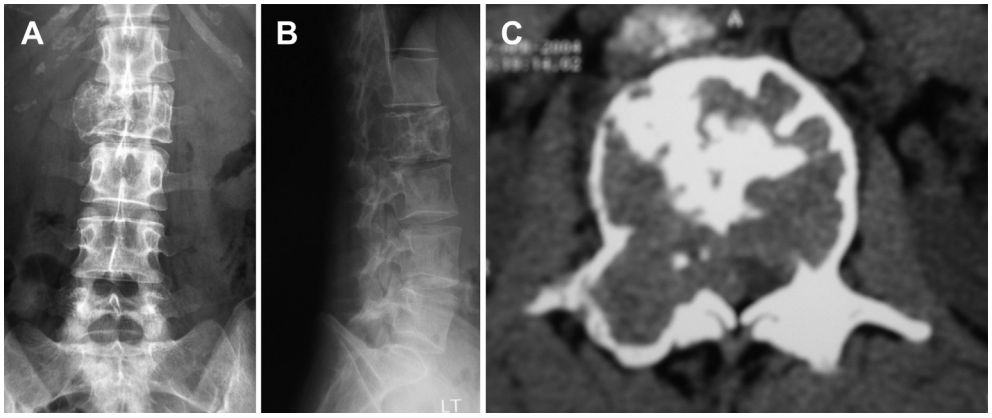

Figure 1. Radiographs of the lumbar spine shows the expansile lesion at L2: (A) anteroposterior and (B) lateral views; (C) computed tomography transverse section of L2.

compression and a provisional diagnosis of GCT was made (Figure 1C). We could not arrange early magnetic resonance imaging (MRI) study of the lumbar spine for this patient.

Intravenous dexamethasone 8 mg three times daily was started immediately. She was advised bed rest with a thoracolumbar jacket. A CT-guided biopsy was done, which showed some proliferation of bland looking spindle shaped cells with oval vesicular nuclei that had occasional nuclear indentations. The nucleoli were inconspicuous with ill-defined cytoplasmic borders. These resembled fibroblasts and histiocytes. There were a few osteoclast-like multinucleated giant cells and thin walled blood vessels. There was no evidence of malignancy but there were focal areas showing degenerated calcifying fibromyxoid tissue. The overall features were more suggestive of an aneurysmal bone cyst (ABC).

Complete intralesional excision, posterior and anterior decompression, instrumentation, and fusion were performed under general anaesthesia. The tumour was found to be friable and greyish white in colour. It was removed piecemeal posteriorly together with the laminae and spinous processes of L1 and L2. L1/L2 and L2/L3 discectomies were done and pedicle screws were inserted two-levels above and two-levels below the lesion with two rods and one cross-link. Fusion was performed with decortication of the laminae and application of artificial bone grafting. An anterior approach was then performed with excision of the left 12th rib. The tumour excision was completed anteriorly and a cage was inserted with bone graft filled as an anterior strut. Postoperatively, dexamethasone was tailed down gradually. Patient controlled analgesia with morphine and oral cyclooxygenase 2 inhibitor were given for pain relief. Perioperative blood loss was about 4000 mL and was replenished with a blood transfusion.

The postoperative period was uneventful. Her neurological status improved immediately after the surgery. By Day 4, she was ambulating well with a brace and a walking frame. The patient was discharged on Day 6. Five weeks later, her wounds had healed well and she had regained full strength of her lower limbs with physiotherapy. Serial X-rays showed evidence of callus formation. The latest X-rays at 2 years showed no loosening of the implants (Figure 2). An MRI scan scheduled 6 months postsurgery revealed no evidence of recurrence (Figure 3). Four years after the operation, she remained asymptomatic.

The final histopathology showed fragments of tumour admixed with trabeculated host bone. The cellular tumour was composed of fairly uniform polyhedral cells displaying mildly pleomorphic and indented (histiocyte-like) nuclei whose mitotic activity was not apparent. Within these tumour fragments were less cellular pinkish myxocartilaginous areas, which also displayed osseous metaplasia and splotchy calcifications. The tumour was also seen juxtaposed to fibrocartilaginous tissue. In addition, multinucleated giant cells were strewn in the tumour. There was no definite evidence of malignant chondroid or osteoid formation seen. The cytomorphological features were compatible with a diagnosis of chondroblastoma of the lumbar vertebra.

Discussion

Primary bone tumours such as chondroblastoma in the spine are rare. Our patient presented to us with a short history and early signs of cauda equina compression resulting from a chondroblastoma of L2.

Three types of chondroblastomas have been described based on their behaviour of growth. The first seldom relapses following curettage and bone grafting, like our patient. The second, more uncommon type has identical histological features but behaves like a malignant tumour with rapid and destructive growth and a tendency to have multiple relapses and metastases.[3] The third and least common type shows malignant transformation.[6] Unfortunately, they cannot be predicted histologically. Close follow-up is required for their final behaviour and course.

Radiologically, chondroblastoma is an osteolytic lesion, sharply delineated from normal bone by a thin rim.[1] There may be mottled densities in the radiolucent zone, depending on calcification of the tumour.[1] One-quarter of these tumours have visible calcification.[7] Cortical destruction is unusual, occurring in 10%.[8] Cortical destruction and extraosseous involvement have been reported in a few patients.[1] These findings may be more common in the vertebral chondroblastomas than in other locations.

Macroscopically, the tissue is grey–blue and elastic–robust. The proliferating cells are oval shaped with well-defined cytoplasmic borders. The nucleus is oval with a longitudinal groove in the middle, creating a coffee bean-like appearance.[1] Benign giant cells are scattered in the lesion. The pericellular, lace-like, fine calcification in the characteristic “chicken wire” or “picket fence” pattern is the hallmark of chondroblastoma.[2] Chondroid differentiation is usually seen. An associated ABC can occasionally (~38%) be found in chondroblastoma, as was seen in our patient.[8] ABCs most commonly appear in the vertebral arch and affect the body secondarily.[9] Pathological diagnosis can sometimes be mistaken for malignancy such as GCT or chondromyxoid fibroma.[10] GCT usually occurs in a skeletally immature patient.[9] GCT has an osteolytic behaviour and spreads from the body to the arch of the vertebrae.[6,9]

With the initial diagnosis of ABC, the recurrence rate is rather high and therefore necessitated total spondylectomy. Although ABCs are indolent and slow growing, surgical decompression was

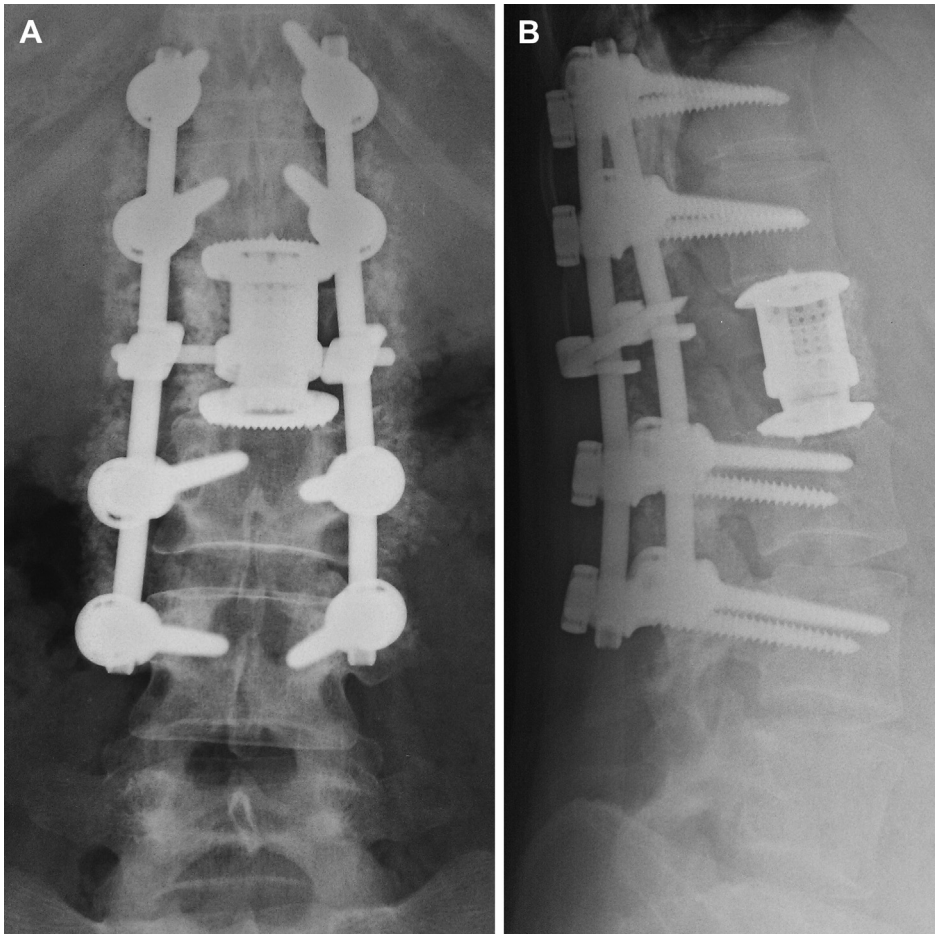

Figure 2. X-rays of the lumbar spine at 2 years postsurgery: (A) anteroposterior and (B) lateral films.

indicated. When the tumour involves the body, pedicles, and part of the lamina, either an anterior or posterior approach alone was insufficient. Combined approaches were required. The lumbar nerves can be preserved. Stabilisation of the spine was achieved with the pedicle screw system posteriorly and the cage system anteriorly in addition to an external brace while awaiting solid fusion. Anterior structural tricortical iliac crest bone graft, can be used but may fail prior to union.[11]

There is a role for preoperative embolisation, especially given the fact that the initial diagnosis was an ABC.[12] This would have

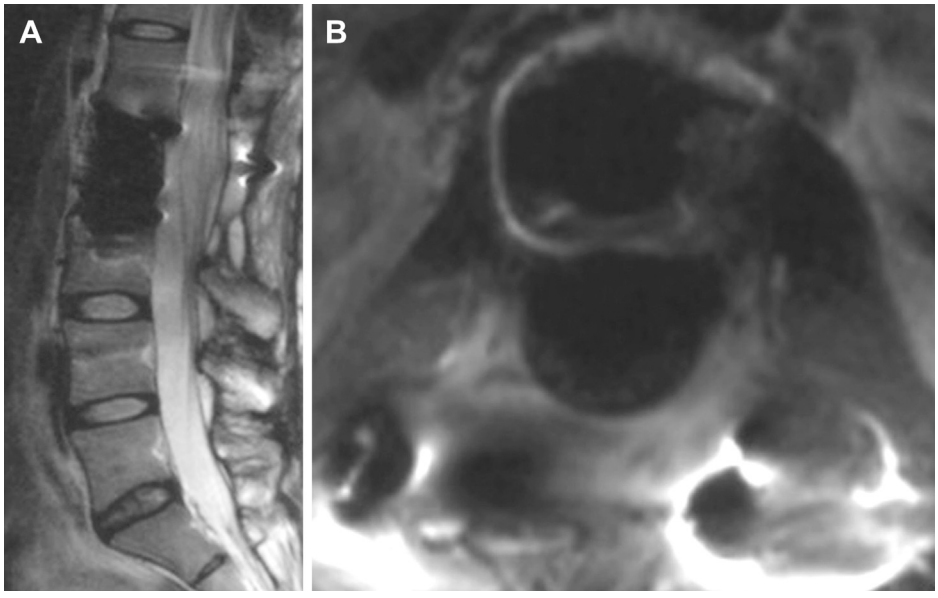

Figure 3. Magnetic resonance images 6 months postoperatively: (A) sagittal and (B) axial views.

helped to reduce perioperative blood loss, but there is a risk to the vascular supply to the spinal cord and cauda equina, which may have further affected her neurological status.

The primary treatment of chondroblastoma is curettage or resection. Curettage cures about 90% of chondroblastomas.[3,6,9] A recurrence rate of about 10% indicates a necessity for complete removal. However, spinal chondroblastomas tend to relapse more frequently, especially if there is an ABC component.[2] Packing of polymethylmethacrylate (bone cement) may decrease the recurrence rate.[5] It can also be treated with repeated curettage and radiation therapy but radiation can increase the risk of sarcomatous transformation. Therefore, it should be used only in recurrences or when surgical clearance is impossible.[6] Chung et al[11] reported that their patient had repeated surgery and radiotherapy but unfortunately succumbed to the disease. There is no role for chemotherapy in the current literature.

There has been one reported case of metastasizing spinal chondroblastoma.[4] It grew more aggressively and invasively than the extraspinal chondroblastoma. It should be treated more radically. They advocated the use of positron emission tomogram CT to detect solid metastases and to resect them in the same setting. Total *en bloc* spondylectomy was probably the best option for our patient.

In conclusion, chondroblastoma of the lumbar spine causing neurological deficit is likely to have a high recurrence rate and aggressive behaviour, therefore, total *en bloc* spondylectomy is the treatment of choice.

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