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A clinical case

Giant Cell Tumor of the L1 Vertebra with a Large Soft Tissue Component: A Case Report

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Abstract

Giant cell tumor (GCT) is a locally aggressive benign tumor that most often appears on long bones. Less than 10% of GCT's in the spine are occur above the sacrum, and cases involving the lumbar spine are relatively little reported in the literature. Soft tissue invasion of GCT is rare.

We present a case of GCT in the L1 vertebra, a large soft tissue component, that occurred in a 24-year-old female patient with low back pain and right radiculopathic thigh pain for approximately 3 months. The patient had no neurological deficit. A lytic lesion involving the L1 vertebral body, more prominently in the right half, extending to the pedicle and transverse process on the right was detected in computed tomography (CT) and magnetic resonance imaging (MRI) sections. In the MR imaging taken 2 months later; It was noted that the mass increased in size and the soft tissue content extending to the right psoas muscle developed. Tru-cut biopsy was performed for the diagnosis and the pathology diagnosis was evaluated as consistent with GCT. The patient underwent L1 vertebral spondylectomy and T1–L3 instrumentation with pedicle screw/mesh cage. Denosumab treatment was started after surgery. The histopathology results of the excised bone and soft tissues were also evaluated as consistent with GCT. As a result, GCT should be considered in the differential diagnosis of destructive lesions with a soft tissue component in the lumbar vertebra.

Key words: giant cell tumor, lumbar spine, soft tissue component, clinical case.

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Introduction

Giant cell bone tumors are rare and constitute approximately 5% of all primary bone tumors (1) and less than 5% of all primary spine tumors [2,3]. These tumors are usually located in the metaphysis or meta-epiphysis of long bones in adults who have completed skeletal maturity between the ages of 20-50. Although GCTs are benign, they are locally aggressive and can metastasize to the lung [4,5]. Spinal GCTs are most commonly located in the sacrum [2,6]. Less than 10% of spinal GCTs is formed above the sacrum [7-9]. GCTs in the sacrum typically occupy more than one segment, while GCTs in the mobile spine are limited to one spine. GCTs originating from the vertebral body cause expansion in the bone structure and pathological fracture by showing pedicle and vertebral posterior extension. Its invasion into adjacent tissues is very rare and there are few reports in the literature [10,11].

In this study, the case of L1 vertebra GCT, a large soft tissue component, is presented and the literature on the subject is reviewed.

Case presentation

A 24-year-old female patient with a history of radicular pain radiating to the lower back and right thigh that had been increasing for about 4 months was admitted to the orthopedics outpatient clinic. The patient had no history of fever, trauma, weight loss and previous infection.

In the physical examination of the patient, pain and tenderness were present when palpated in the lumbar region. Lumbar movements were painful and restricted. No pathological findings were detected on neurological examination. Laboratory tests were within normal limits. In the radiographs of the patient taken in an external center two months ago; An expansile lytic lesion was observed in the L1 vertebral corpus (Figure 1).

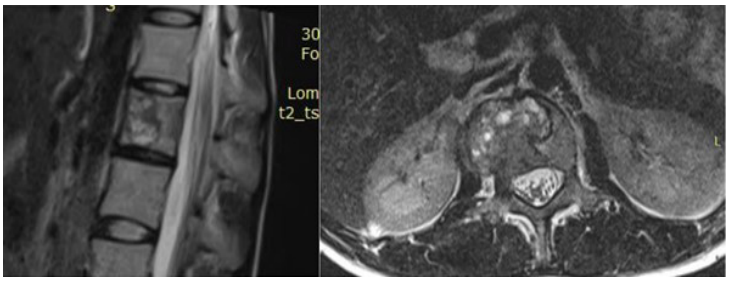

Figure 1 - Patient's first sagittal and axial MR images

A lumbar vertebral CT examination was requested for a more detailed evaluation. On CT, an extrapanile lytic lesion was observed, which largely covers the vertebral corpus, mainly in the right half, and extends to the pedicle and transverse process on the right. There was a soft tissue component that filled the anterior epidural fat distance of the lesion and extended to the paravertebral area on the right. Lumbar vertebra MR examination was requested to better evaluate the spinal canal. In lumbar spine MRI, it was noted that the lesion had a heterogeneous solid structure containing millimetric cystic areas in T2W, hypo-intermediate signal feature in T1W and intense contrast in T1W postcontrast sections. It was observed that the soft tissue component filling the anterior epidural fat space of the mass lesion pressed the thecal sac and the nerve roots on the right. The lesion had a large soft tissue component extending into the right paravertebral area and into the psoas muscle (Figure 2, 3).

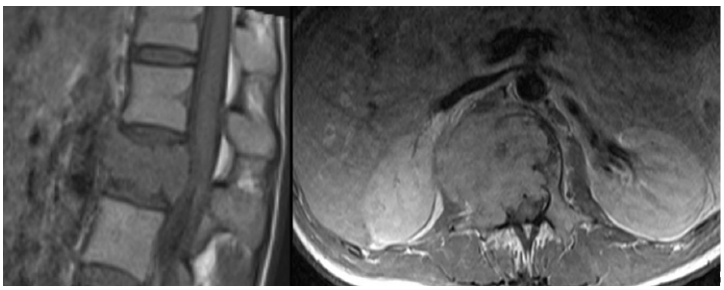

Figure 2 - Sagittal and axial MR images of the patient two months later

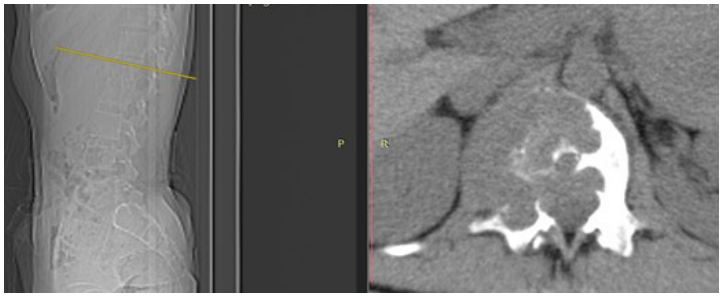

Figure 3 - Sagittal and axial CT images of the patient two months later

A tru-cut biopsy was performed on the patient to confirm the diagnosis. The biopsy result was evaluated as consistent with DHT (Figure 4).

Due to the development of a pathological fracture secondary to a giant cell tumor in the L1 vertebra, the patient was initially planned for vertebral stabilization with tumor excision, and denosumab treatment was started.

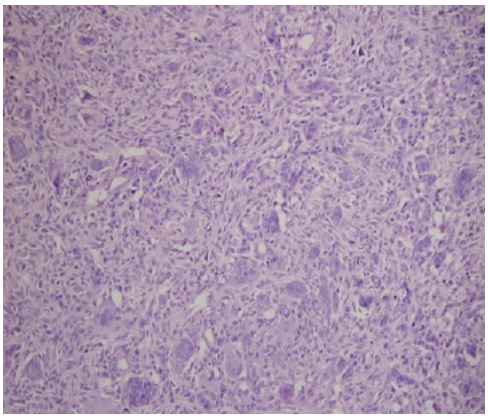

Figure 4 - Numerous uniformly distributed giant cells in a spindle cell stroma without atypia. x 200 Hematoxylin-Eosin

The surgical procedure was performed in the prone position with the posterior intervention. A long posterior incision was made along the T11 and L3 vertebrae, centering the L1 vertebra. In the opening of the surgical field, the spine was exposed by preserving the supraspinous and interspinous ligaments, the cranial and caudal facet joint capsules. Pedicle screws were inserted into the T11, T12, L2 and L3 vertebrae using the free hand technique. Then, the tumor in the L1 vertebra and the psoas muscle was reconstructed by spondylectomy, wide resection and mesh cage. The removed tumor materials were sent for histopathological examination. No neurological symptoms were observed in the patient postoperatively. We started treating the patient with denosumab 120 mg once a month.

Discussion

The most common complaints of patients with spinal GCT are low back and radicular thigh pain [12,13]. Patients are usually diagnosed with GCT after developing pain or neurological deficits due to a pathological fracture [14,15].

Spinal GCT is not common and constitutes approximately 16.2% of all primary spinal tumors [16]. Spinal GCTs are most commonly seen in the sacrum, then in order of frequency in the thoracic, cervical and lumbar spines, [17] It is more common in women than men [18]. It is usually seen in adults who have completed their skeletal maturity, most commonly in the third decade [19]. While GCT of more than one sacral segment is common in the sacral part of the spine, invasion of GCT into adjacent bones and soft tissues in other parts of the spine is rare [10,11,18]. The age of the presented case is a suitable age for GCT. It formed an osteolytic lesion in the spine in a short period of two months, and it invaded the right psoas muscle and caused the muscle to expand, and specific infections of the spine should be excluded in the differential diagnosis in terms of such a radiological appearance. Invasion to adjacent segment spine and soft tissues is rare in spinal GCTs [20-22], and Yuan B. et al. named cases with such behavior as atypical GCTs of the spine [21]. However, the invasion of the surrounding tissues in a short period of time, as in the case we presented, has not been reported in the literature.

It is generally recommended to exclude aneurysmal bone cyst, osteoblastoma and plasmacytoma of the spine in the differential diagnosis of spinal GCTs [10,20]. In the presented case, it was clinically and radiologically consistent with a specific infection of the spine. Therefore, we think that the specific infection of the spine, tuberculosis spondylodiscitis, should also be excluded in the differential diagnosis. In the presented case, similar to the characteristic clinical features of tuberculous spondylodiscitis, there was local pain in the back, tenderness, spasm in the paraspinal muscles and a prominent gibbus deformity in the back region. Radiologically, vertebral body collapse, local kyphotic deformity and psoas abscess-like appearance were present [23,24].

Wide en bloc resection is recommended in GCTs, especially in Enneking 3rd stage [25]. However, a wide resection of the spine is not always possible due to adjacent anatomical structures [26]. Local recurrence rate of GCTs has been reported as 27-65% in curettage alone, 12-27% in curettage combined with adjuvant therapy, and 0-12% in en-bloc resection [27-30]. Different adjuvant treatments are available to reduce the recurrence rate of GCT. In recent years, good results have been reported regarding Denosumab therapy in the treatment of GCT and in preventing its recurrence [31].

Osteoclast-like giant cells express receptor activator of nuclear factor-kappa B(RANK); stromal cells, the neoplastic component of GCT, express RANK ligand (RANKL), and RANKL provides the formation of osteoclast [32,33]. Excessive RANKL release is associated with GCT [34]. Denosumab en block, a fully human monoclonal antibody that inhibits RANKL, is a promising treatment for unresectable spinal GCTs [35,36], and has been shown to be a potentially beneficial treatment for spinal GCTs, including the sacrum [31].

There are many studies showing that the recurrence rate is high (40-45%) if good resection of the tumor tissue is not performed despite preoperative administration of denosumab [37-39]. Li H et al. reported that the reason for the high recurrence rate in patients treated with preoperative denosumab was that the thickened new bone induced by denosumab and entrapped with tumor cells makes it difficult for the surgeon to identify the true size of the tumor and perform an adequate curettage [40].

In their study, Yayama T et al. suggested that denosumab does not completely eliminate GCT cells, and therefore, denosumab treatment should be started after an effective surgical resection of the tumor [41]. In our case, we started denosumab treatment after total spondylectomy and extensive soft tissue resection.

Conclusions

Tissue biopsy is the gold standard for the diagnosis of spinal GCT. Generally, in the differential diagnosis of spinal GCTs, it is recommended to exclude aneurysmal bone cyst, osteoblastoma and plasmacytoma of the spine due to their localization and radiological appearance in the spine. However, as in our case, we think that due to the atypical behavior of GCTs in the spine, specific infections of the spine should also be excluded in the differential diagnosis.

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L1 омыртқасының жұмсақ тіндік үлкен компоненті бар алып жасушалы ісігі: Клиникалық жағдай

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Түйіндеме

Алып жасушалы ісік (АЖІ) – көбіне ұзын сүйектерде орналасатын жергілікті агрессивті қатерсіз ісік. Омыртқадағы АЖІ-тің 10%-дан азы құйымшақтың үстінде болады, ал бел омыртқасында кездесетін жағдайлар әдебиетте салыстырмалы түрде аз жарияланған. Сонымен қатар АЖІ -тің жұмсақ тіндік инвазиясы сирек кездеседі.

Біз 24 жастағы әйел науқастың L1 омыртқасындағы жұмсақ тіндердің үлкен компонентімен анықталған АЖІ клиникалық жағдайын ұсынамыз. Науқасты 3 ай бойы бел аймағындағы ауырсыну мен оң жақ сан аймағындағы радикулопатиялық ауырсыну мазалаған. Неврологиялық дефицит орын талмаған. Компьютерлік томография (КТ) және магнитті-резонанстық томография (МРТ) нәтижелерінде L1 омыртқа денесінің оң жақ аяқша мен көлденең өсіндіге дейін тараған литикалық зақымдануы анықталды. Ал 2 ай өткенде түсірілген МРТ нәтижесінде масса мөлшерінің ұлғайғанын және белдің оң жақ бұлшықетіне дейін таралған жұмсақ тіндік компонентті анықтады. Tru-cut биопсиясы қорытындысы бойынша патология диагнозы АЖІ ретінде танылды. Науқасқа L1 омыртқасының спондилэктомиясы және T1–L3 омыртқаларын транспедикулярлы винт/торлы кейдж көмегімен тазарту жүргізілді. Отадан кейін деносумабпен ем жүргізілді.

Кесілген сүйек пен жұмсақ тіндерді гистологиялық зерттеу нәтижелері де АЖІ диагнозы растады. Осы тәжірибемізді ескере отырып, біз бел омыртқасының жұмсақ тіндік компоненті бар деструктивті зақымданулардың ажыратпалы диагностикасында АЖІ ескерілуі керек деп санаймыз.

Түйін сөздер: алып жасушалы ісік, бел омыртқасы, жұмсақ тіндік компонент, клиникалық жағдай.

Гигантоклеточная опухоль позвонка L1 с крупным мягкотканым компонентом: Клинический случай

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Резюме

Гигантоклеточная опухоль (ГКО) — локально агрессивная доброкачественная опухоль, чаще всего локализуются в длинных костях. Менее 10% ГКО в позвоночнике встречаются выше крестца, а случаи, поражающие поясничный отдел позвоночника, относительно мало описаны в литературе. Инвазия мягких тканей ГКО встречается редко.

Мы представляем случай ГКО в позвонке L1 у 24-летней пациентки, в большом компоненте мягких тканей. Пациента беспокоили боли в пояснице и радикулопатические боли в правом бедре в течение 3 месяцев. У пациента не было неврологического дефицита. На срезах компьютерной томографии (КТ) и магнитно-резонансной томографии (МРТ) было обнаружено литическое поражение тела L1 позвонка, распространяющееся на ножку и поперечный отросток справа. На МРТ, сделанном через 2 месяца, было отмечено, что масса увеличилась в размерах и развилось мягкотканное содержимое, распространяющееся на правую поясничную мышцу. Для диагностики была проведена биопсия Tru-cut, и диагноз патологии был оценен как ГКО. Пациенту была выполнена спондилэктомия позвонка L1 и инструментальная обработка T1–L3 с помощью транспедикулярного винта/сетчатого кейджа. Лечение деносумабом было начато после операции. Результаты гистопатологии иссеченной кости и мягких тканей также были оценены как соответствующие ГКО. В связи с этим ГКО следует учитывать при дифференциальной диагностике деструктивных поражений с мягкотканым компонентом поясничного позвонка.

Ключевые слова: гигантоклеточная опухоль, поясничный отдел позвоночника, мягкотканый компонент, клинический случай.