Clinical Case Report

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Lumbar spinal canal osteosarcoma

A case report

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Abstract

Rationale: Osteosarcoma is a rare neoplasm in the lumbar spine. Although osteosarcoma can arise in any portion of the skeleton, it very rarely arises in the spinal canal, which accounts for <0.1% of all cases of adult sarcomas. Here, we describe a case of osteosarcoma arising in the L4–5 spinal canal.

Patient concerns: The present report describes the case of a 55-year-old female patient with osteosarcoma of the L4–5 spinal canal.

Diagnoses: The patient was initially diagnosed with lumbar spinal stenosis and underwent lumbar fusion at a local hospital. At the 4-month follow-up, the patient reported a marked increase in numbness and pain in the lumbar region and lower limbs. Based on magnetic resonance imaging, we diagnosed a postoperative infectious lesion of the lumbar spine.

Interventions: The patient underwent surgery for complete removal of the mass lesion. The mass measured $3 \times 2.5 \times 0.7\,cm^3$ in size and was located in the L4–5 spinal canal.

Outcomes: Based on histological and immunohistochemical findings, the diagnosis of osteosarcoma was confirmed by an expert pathology consultant. The patient then received chemotherapy. Postoperative follow-up at 6 months revealed no evidence of recurrent disease or residual side effects from therapy.

Lessons: Osteosarcoma in the L4–5 spinal canal is extremely rare and very difficult to distinguish histologically from benign nervous and fibrous tissue. This is a very valuable case, which highlights the need for orthopedic surgeons to consider this when diagnosing patients with spinal tumors.

Abbreviations: ADM = doxorubicin, CT = computed tomography, DDP = cisplatin, HE = hematoxylin and eosin, IFO = isophosphate, MRI = magnetic resonance imaging.

Keywords: imaging, immunohistochemistry, lumbar spinal canal, misdiagnosis, osteosarcoma, pathology, surgery

1. Introduction

Osteosarcoma is a primary malignant bone tumor originating from stem cells and is characterized by proliferation of tumor cells that directly form immature bone or bone-like tissue. Osteosarcoma most commonly occurs in young people and has an incidence rate of approximately 0.3/million, accounting for an estimated 0.2% of all malignant tumors.[1] Osteosarcoma exhibits a predilection to occur in the metaphysis of long bones, and most commonly occurs in the distal femur (43%), proximal tibia (23%), or humerus (10%).[2] Osteosarcoma represents only 3% to 5% of all spinal malignancies.[3,4] The sacral area, followed by the lumbar and thoracic spine segments, are the most common locations.[5] Osteosarcoma is a rare neoplasm in the lumbar spinal canal, and the probability of osteosarcoma occurring as a spinal tumor is extremely low.[6] There is very little research on spinal canal osteosarcoma and thus limited experience in the diagnosis of osteosarcoma in the spinal canal after lumbar fusion. Thus, this type of osteosarcoma is very easy to misdiagnose and difficult to distinguish from postoperative infectious lesions and thrombi.

Osteosarcoma is diagnosed on the basis of clinical and laboratory examination, imaging analysis, and histopathologic studies. However, the clinical symptoms, laboratory data, and imaging findings are not specific and are similar to those seen with lumbar disc herniation or neurofibroma. Histopathologic analysis with immunohistochemical examination are diagnostic requirements for osteosarcoma. Thus, osteosarcoma of the spinal canal is easily misdiagnosed before surgical removal of the lesion. Here, we present a case of osteosarcoma of the spinal cord that was misdiagnosed as an infectious lesion following surgery. This misdiagnosis resulted in progression of the mass and inadequate treatment, and only after resection of the lesion and histological and immunohistochemical analysis was the diagnosis of

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osteosarcoma confirmed. The present study was approved by the Ethics Review Committee of The First Affiliated Hospital of Nanchang University (Nanchang, China), and written informed consent was obtained from the patient.

2. Case presentation

In October 2016, a 54-year-old female patient presented to the Orthopedic Clinic of Leping County People's Hospital (Jiangxi, China) with a chief complaint of low back pain with pain and numbness of the right lower limb for 4 months. During this period, the patient was treated with conservative management including physiotherapy and spinal steroid injections, which had no obvious curative effect. The local district general hospital radiologist analyzed digital radiography (Fig. 1) and computed tomography (CT) (Fig. 2) and diagnosed lumbar disc herniation and L4 spondylolisthesis. The patient was advised to undergo L4/5 posterior decompression and fusion surgery (Fig. 3). However, 4 months later, in February 2017, the patient reported a gradual increase in lumbar and right limb pain. The patient was referred to the Department of Orthopedics of The First Affiliated Hospital of Nanchang University for further treatment. Personal and family histories were significant for a right femoral neck fracture and artificial hip arthroplasty 14 years prior. General physical examination demonstrated that passive and active range of motion of the limbs was normal, with the exception of paresthesia in the right lower limb. No fever or respiratory compromise was noted, and no history of weight loss or exposure to tuberculosis was reported by the patient. Additionally, physical examination revealed no palpable head, neck, supraclavicular, axillary, or epitrochlear lymph nodes. Inflammatory markers, including erythrocyte sedimentation rate and C-reactive protein, were within the normal ranges. After whole body examination, there were no primary tumor lesions identified.

Magnetic resonance imaging (MRI) was performed for further evaluation. Sagittal T1-weighted images (Fig. 4A) revealed a mass-like lesion on the spinal signal. Sagittal T2 and fat-suppressed T2-weighted images (Fig. 4B and C) showed high signal lesions near the spinal cord and axial T2-weighted images (Fig. 4D) also demonstrated high-signal lesions compressing the spinal cord in the L4–5 spinal canal. Physicians in the department of imaging diagnosed postoperative infectious lesions of the lumbar spine. However, the authors were skeptical of this diagnosis. Therefore, the authors decided to perform surgical exploration and complete excision of the lesion for pathological examination. The patient had no surgical contraindications, and surgeons specializing in spine disease performed the surgery. The mass was identified and en bloc excision was performed. The lesion originated from an operative scar from previous surgery and was located in the low part of the left vertebral arch of the 4th lumbar vertebra; approximately 2/3 of the tumor was located in the spinal canal, compressing the dural sac (Fig. 5). The lesion was medium-sized with a rough surface and appeared similar to a sarcomatous mass upon gross examination; it measured approximately $3 \times 2.5 \times 0.7$ cm^3 in size. Hematoxylin and eosin (HE) and immunohistochemistry examination of the lesion was performed at The Affiliated Tumor Hospital of Shanghai Fudan University. HE examination showed tumor cells that were flaky, bulky, and polygonal with rich red staining cytoplasm, large nuclei, visible nucleoli, a mitotic rate >20/10 high power fields, and interstitial bone-like matrix; in some areas, sparse star-shaped tumor cells were present with fibrous tissue and fibrous changes. Immunohistochemistry results were as follows: actin negative, CD10 <10% positive, CDX2 negative, CEA negative, CK20 negative, CK7 negative, desmin negative, estrogen receptor negative, GFAP negative, Ki-67 positive in 50% +, P120 positive, P63 positive, progesterone receptor positive, S100 negative, villin negative, vimentin 2+ positive, and smooth muscle actin negative. Based on these examinations, the patient was diagnosed with osteosarcoma (Fig. 6).

After a definitive diagnosis, she was administered 6 cycles of systemic intravenous chemotherapy consisting of cisplatin (DDP) 120 mg + 0.9% NaCl 500 mL, ivgtt/qd, for 1 day, and doxorubicin (ADM) 20 mg + 0.9% NaCl 250 mL, ivgtt/qd for 3 days. After 2 weeks, isophosphate (IFO) 2 g + 0.9% NaCl 500 mL, ivgtt/qd were administered for 5 days. After 2 weeks, the cycle was repeated. After 6 cycles of systemic intravenous chemotherapy, the patient was discharged without any complications. At the 6-month follow up, consisting of plain radiography and MRI, the patient was symptom-free and able to return to work. At present,

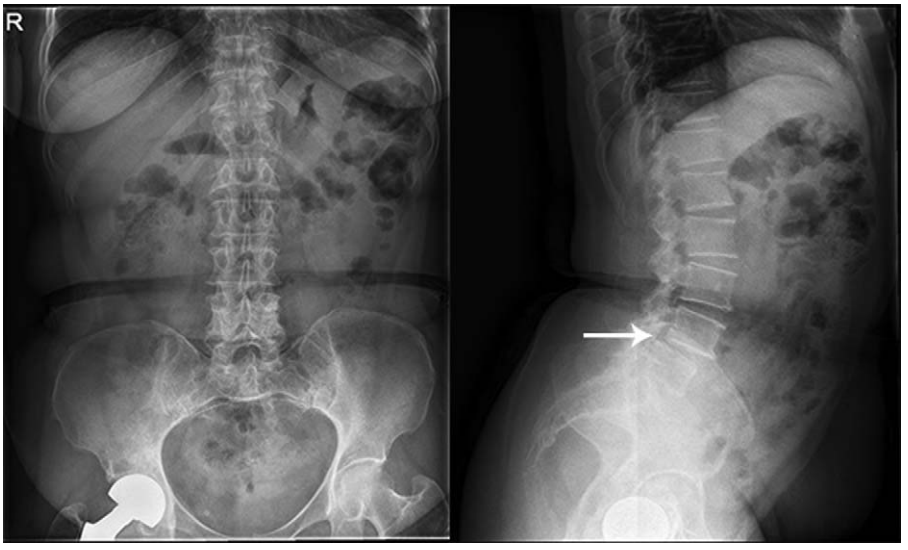

Figure 1. Digital radiography showed L4 vertebral spondylolisthesis and right hip prosthesis.

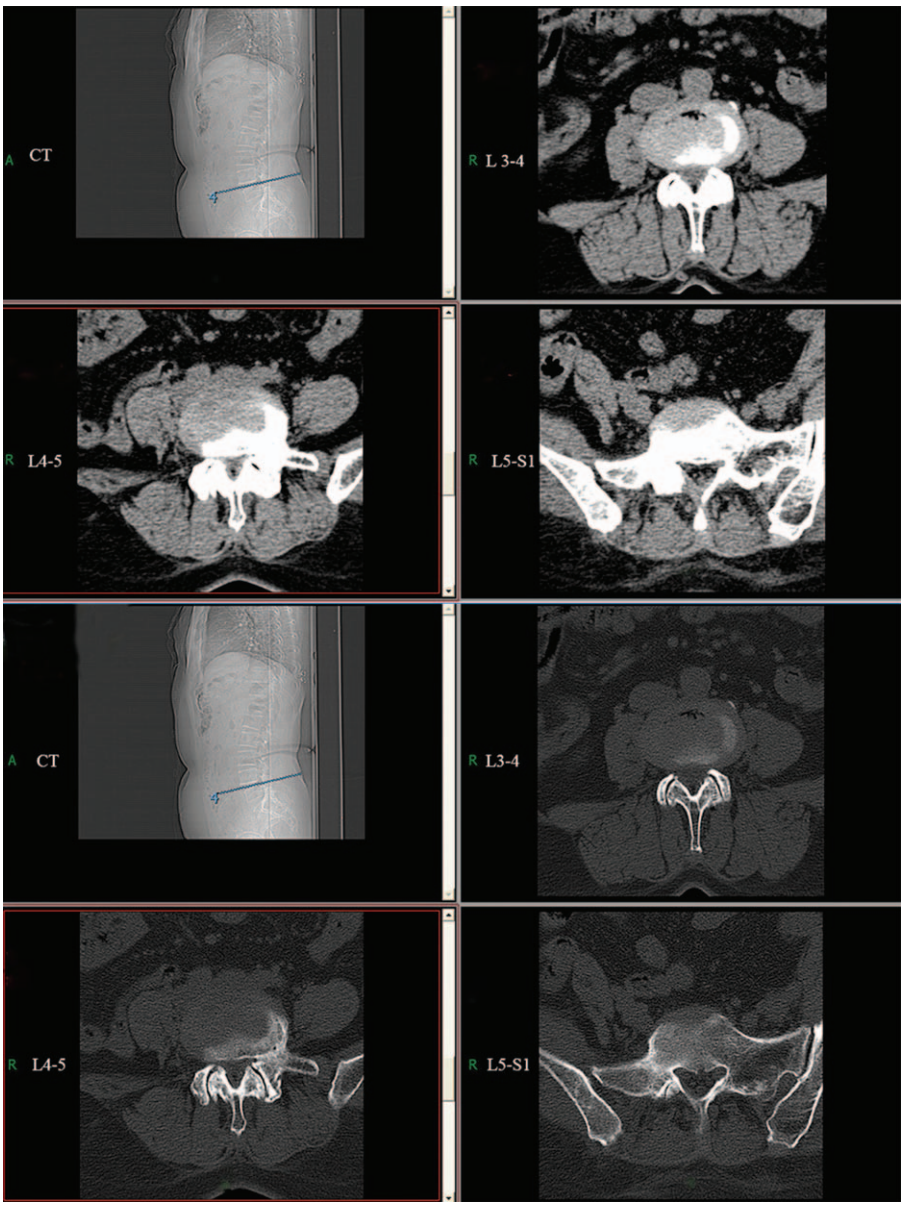

Figure 2. Computed tomography revealed spinal stenosis and L4 spondylolisthesis. Abnormal mass lesions were not found.

the patient is currently alive and well. However, close monitoring is required in this case due to the high rate of recurrence and metastasis associated with osteosarcoma.

3. Discussion

Lumbar canal osteosarcoma is very rare. Osteosarcoma occurs most frequently in the appendicular skeleton, especially the distal femur and proximal tibial metaphysis, which account for 90% of all cases.[3,4] Primary osteosarcomas of the vertebral column are uncommon; to our knowledge, a total of 78 cases, most located in the vertebral body, have been previously reported.[7] However, osteosarcoma in the lumbar canal arising from the postoperative field has not been reported in the previous literature. As for extraskeletal osteosarcoma, according to Terence's study, the location of the tumor has a history of trauma, which accounts for 12.5% and 30.7%, and the history of radiation therapy is 5.7% and 10%.[8] In the present study, we report a case of primary tumor arising in a postoperative field, which was misdiagnosed with infectious lesions and led to progression of the disease; therefore, MRI is crucial for the diagnosis of soft tissue masses. We thought that MRI should be performed before lumbar fusion to determine if other lesions were the cause of lower back pain in the patient. Although MRI cannot definitively diagnose a mass lesion, the authors consider MRI an important imaging examination for early detection of osteosarcoma in the spinal canal.

In this case, MRI was performed for the evaluation of the lesion. Sagittal T1 images (Fig. 4A) revealed an abnormal elongated T1 signal on the left spinal canal at the L4/5 intervertebral space. Axial T2-weighted images (Fig. 4B) revealed an abnormally shaped, slightly elongated T2 signal at the left spinal canal L4/5 intervertebral space. Fat-suppressed T2-weighted images (Fig. 4C) also demonstrated the lesion with a high signal, compressing the adjacent dural sac. Radiology physicians believed that the masses were postoperative infection lesions, based on MRI. However, the patient had no symptoms of infection, such as

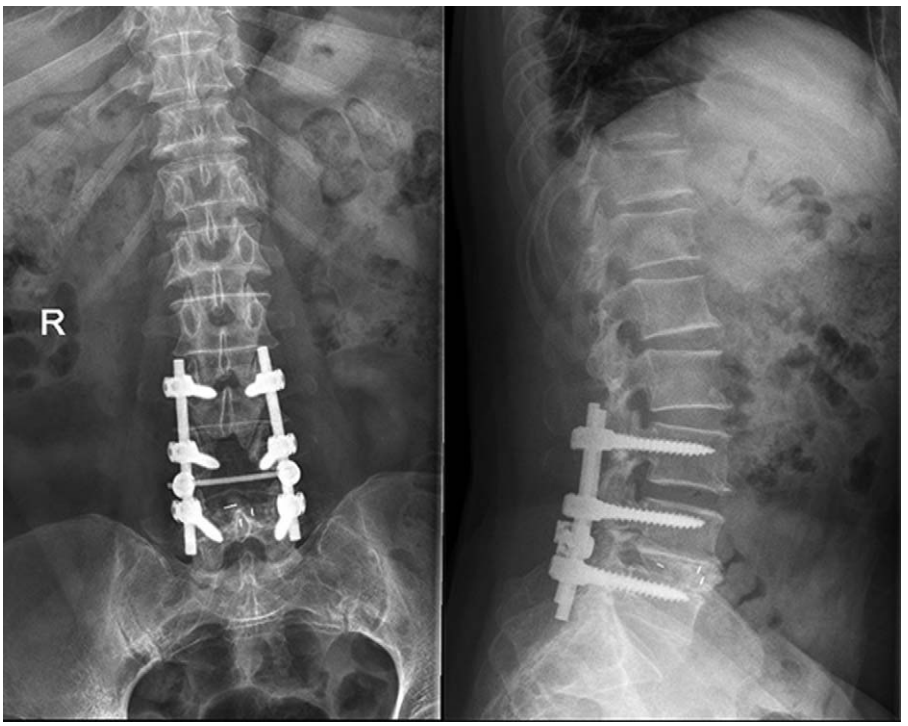

Figure 3. Postoperative digital radiography after L4 vertebral spondylolisthesis reduction and internal fixation and lumbar 4–5 fusion, showing better position of the nail bar and the fusion device.

fever or wound exudate. The authors questioned the diagnosis of infection because infectious lesion signals were similar to muscle tissue on T1-weighted images, and the boundary was unclear in lesions involving the entire vertebral level. Therefore, the authors believed that the lesion was not infectious in nature. Additionally, we could also exclude neuronal-derived tumors because neurogenic tumor signal is the same or slightly lower than that of the dura mater on the T1-weighted images and the signal increases on T2-weighted images. Therefore, MRI is an important means of identifying lesions in the spinal canal. Finally, the authors advocated surgical removal of the lesion for pathological examination and definitive diagnosis.

In this difficult case, pathology with immunohistochemistry was crucial due to the variable prognoses and therapeutic approaches used to treat infection lesion as opposed to neoplastic tumors. Pathologic examination with immunohistochemistry is considered valuable for the diagnosis of infection or tumors, as a variety of markers with a range of specificity and sensitivity are available.[9,10] The histopathological diagnosis is based on morphological, immunohistochemical, and ultrastructural findings revealing a skeletal muscle phenotype.[11] Histology can distinguish postoperative inflammation, healing bone, early fusion, bone graft material, and possibly superimposed infection. Based on pathologic examination including immunohistochem-

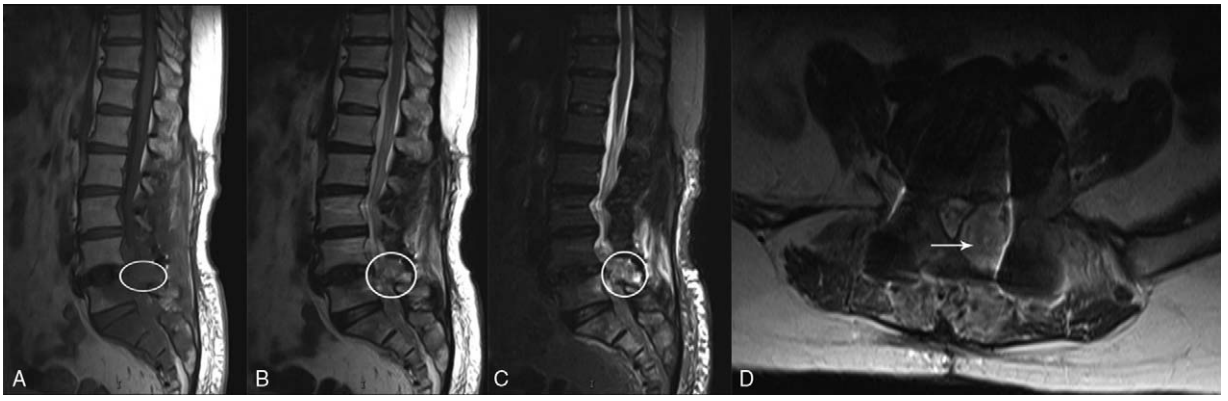

Figure 4. (A) Sagittal T1 magnetic resonance imaging (MRI) revealed an abnormal elongated lesion on the left side spinal canal at the level of the L4/5 intervertebral space. (B) Axial T2-weighted MRI showed an abnormally shaped, slightly longer lesion on the left side of the spinal canal at the L4/5 intervertebral space. (C) Fat-suppressed T2-weighted MRI showed the lesion with a high signal, compressing the adjacent dural sac. (D) Transverse MRI showed abnormal tissue compressing the spinal cord, with a signal similar to that of the spinal cord.

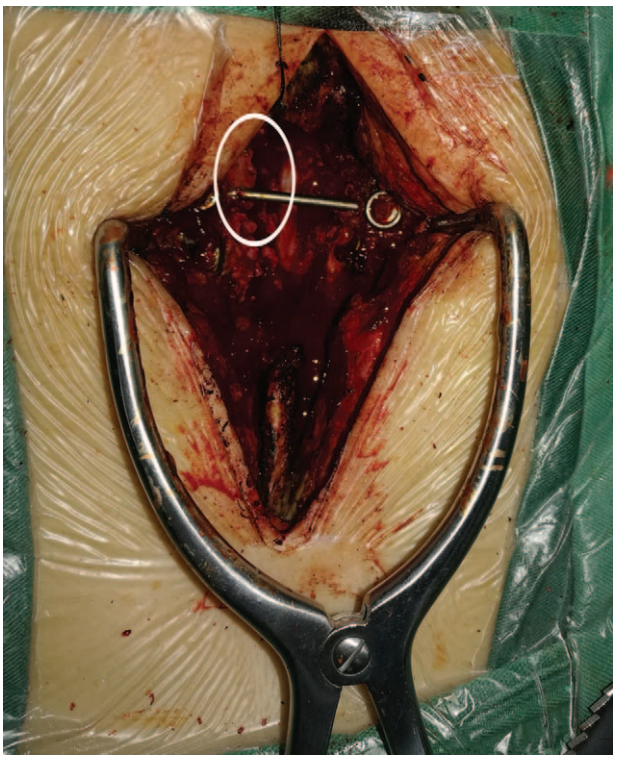

Figure 5. Intraoperative image. The mass is located in the L4–5 spinal canal, compressing the spinal cord. The mass was completely resected.

istry, the diagnosis of osteosarcoma was made. This represents an extremely rare case of osteosarcoma of the lumbar spinal canal, a lesion that is very easy to misdiagnosis.

Surgical excision of osteosarcoma in the spinal canal is considered as the preferred treatment, since it relieves spinal cord compression and allows for confirmation of the diagnosis pathologically. The most effective surgical intervention for spinal osteosarcoma is wide, en bloc resection, which is defined as removal of the tumor in a single piece, with a surrounded rim of healthy tissue outside the pseudocapsule.[12,13] Additionally, osteosarcoma is sensitive to chemotherapy; thus, complete necrosis of tumor cell may be achieved with chemotherapy. Ferrari and Serra[14] reported that osteosarcoma has a high response rate to chemotherapy, especially neoadjuvant chemotherapy. With the emergence of neoadjuvant chemotherapy, the 5-year survival rate of patients with osteosarcoma has increased to 60% to 70%.[15] Therefore, the authors suggest that osteosarcoma of the spinal canal must be treated with neoadjuvant chemotherapy followed by en bloc resection, to achieve complete necrosis of tumor cells and prevention of recurrence. However, histopathological examination and neoadjuvant chemotherapy were not performed before surgery in the patient mentioned in this report. Our study was limited because we misdiagnosed the lesion as lumbar infection before surgery.

In conclusion, the present case describes a 55-year-old female patient who suffered from primary osteosarcoma of the spinal canal. MRI, pathologic examination including immunohistochemistry, and en bloc excision were performed, and at 6-month follow-up, the patient had no evidence of recurrent disease or residual side effects from therapy. The performance of MRI and pathologic evaluation with immunohistochemistry is particularly important for the differential diagnosis of soft tissue tumors. The follow-up time of the current patient is relatively short considering the high risk of recurrence and metastasis associated with misdiagnosed osteosarcoma of the spinal canal, and long-term follow-up is often recommended in such cases. As

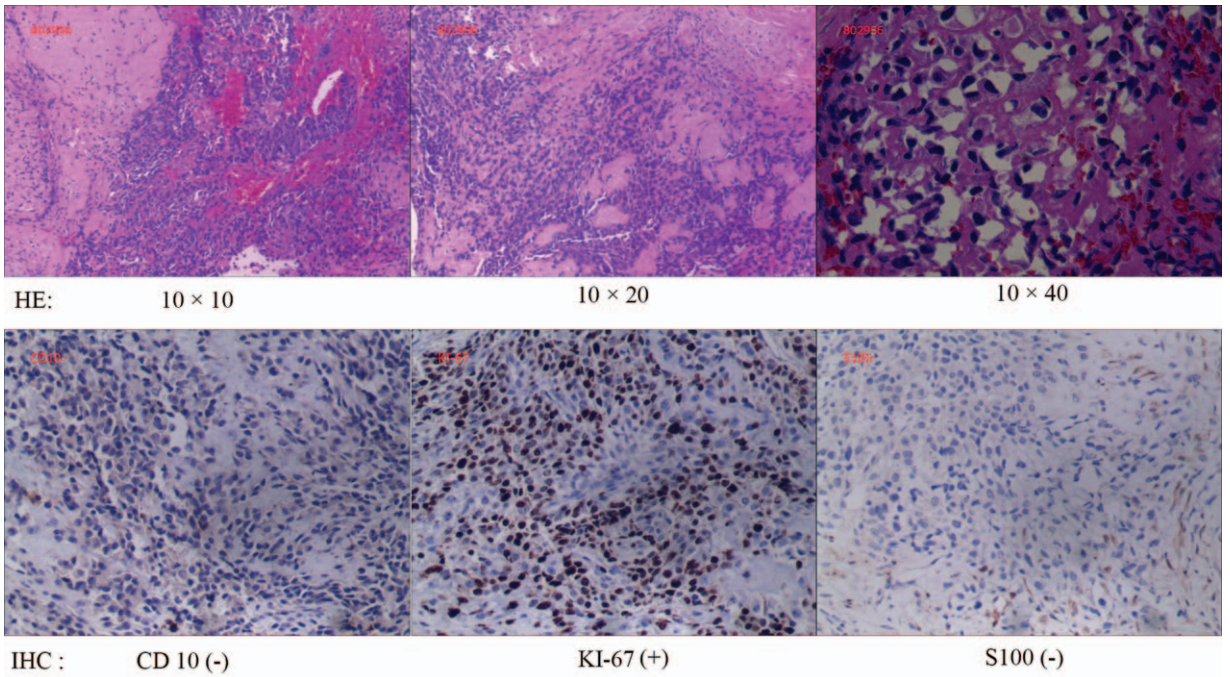

Figure 6. Histopathology (hematoxylin and eosin) showed tumor cells that were flaky, bulky, and polygonal, with rich red staining cytoplasm, nuclear enlargement, visible nucleoli, a mitotic rate >20/10 high power fields, and interstitial bone-like matrix in some areas; rare tumor cells were star-shaped with adjacent fibrous tissue and fibrous changes. Immunohistochemistry results were as follows: actin(−), CD10 (<10% +), CDX2(−), CEA(−), CK20(−), CK7(−), Des(−), ER(−), GFAP(−), Ki-67(50%+), P120(+), P63(−), PR(−), S100(−), villin(−), Vim(2+), and SMA(−).

osteosarcoma of the spine has a poor prognosis, 20% of patients develop local recurrence after en bloc excision, compared to 60% patients who develop after intraregional excision.[16,17]

Author contributions

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Writing – review & editing: De-jian Chen.

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