

Osteosarcoma with Pathological Fracture: A Rare Case Report

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ABSTRACT

Introduction: Osteosarcoma is a primary malignant bone tumor originating from stem cells and is characterized by the proliferation of tumor cells that directly form immature bone or bone-like tissue. Osteosarcoma represents only 3% to 5% of all spinal malignancies. The aim of study is to discuss the importance of diagnostic investigation for a relatively rare case of spinal osteosarcoma.

Case presentation: A 49 years old female came to the Emergency Unit, with a chief complaint of bilateral weakness of lower extremities for 1 year and worsening in 2 weeks. Upon examination, the initial diagnosis was mistaken for a spinal canal stenosis because metastatic bone disease from previous history of malignant melanoma. Subsequently, a decompression with stabilization fusion and biopsy is performed. Primary osteosarcoma was confirmed from histopathology results. Further chemotherapy is planned referring to confirmed primary osteosarcoma on histopathology.

Discussion: Spinal 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. Primary osteosarcomas of the vertebral column are uncommon and occur dominantly in the

vertebral body. Biopsy remains the gold standard of care for diagnosis.

Conclusion: The performance of MRI and pathologic evaluation with immunohistochemistry is particularly important for the differential diagnosis of soft tissue tumors. Histopathologic analysis also plays an important role in differentiating from metastatic bone disease. An aggressive treatment plan and long-term follow-up are mandatory in managing this case.

Keywords: osteosarcoma, spinal osteosarcoma, spinal canal stenosis, malignancy, pathological fracture, rare case.

INTRODUCTION

Osteosarcoma is a primary malignant bone tumor originating from stem cells and is characterized by the 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 occurs 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 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 the immunohistochemical examination is a diagnostic requirement 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 spine that was misdiagnosed with metastatic bone disease from previous history of malignant melanoma. And only after decompression with stabilization fusion and biopsy is performed, histological analysis of the biopsy was confirmed with osteosarcoma. The aim of the study is to discuss the importance of diagnostic investigation for a relatively rare case of spinal osteosarcoma.

CASE REPORT

A 49-year-old female came to the Emergency Unit of Prof. Dr. IGNG Ngoerah Hospital, with a chief complaint of bilateral

weakness of lower extremities for 1 year and worsening 2 weeks. The patient also had low back pain. There is a past history of malignant melanoma on the right axilla (based on biopsy examinations). On local physical examinations, midline tenderness at levels L3-S1 with hypoesthesia at a level below L1 (Fig. 1), pathologist reflex such as Babinski was positive.

Laboratory results revealed increasing alkaline phosphatase (ALP) 909 U/L, Erythrocyte sedimentation rate (ESR) 50 mm/hour. Meanwhile, on imaging examinations such as Magnetic resonance imaging (MRI) showed malalignment with kyphotic deformity at levels Thoracic 9 – 10. Suggestive of metastatic process on vertebral corpus at levels C5, Thoracic 4 - 5, Thoracic 10-12, S3 – S5, with compression of pathological fracture at level Thoracic 10. (Fig 2 – 5). Meanwhile, myelography examination showed partial liquor cerebrospinal fluid at level Thoracic 10 (Fig 6). These results concluded that malignancy at the level of Thoracic 10 with spinal canal stenosis grade III.

After the final evaluation, we informed consent to the patient and decided to perform decompression with stabilization fusion and biopsy examination (Fig 7-8). After the surgery was done, the patient applied a thoracolumbar sacral orthosis (TLSO) brace following rehabilitation protocol. Based on immunohistochemistry biopsy confirmed osteosarcoma with fibroblastic conventional in region pedicle and lamina at level Thoracic 11 - 12. The patient was scheduled for chemotherapy in an oncology outpatient clinic.



Figure 1. Physical examination of back pain (posterior views)

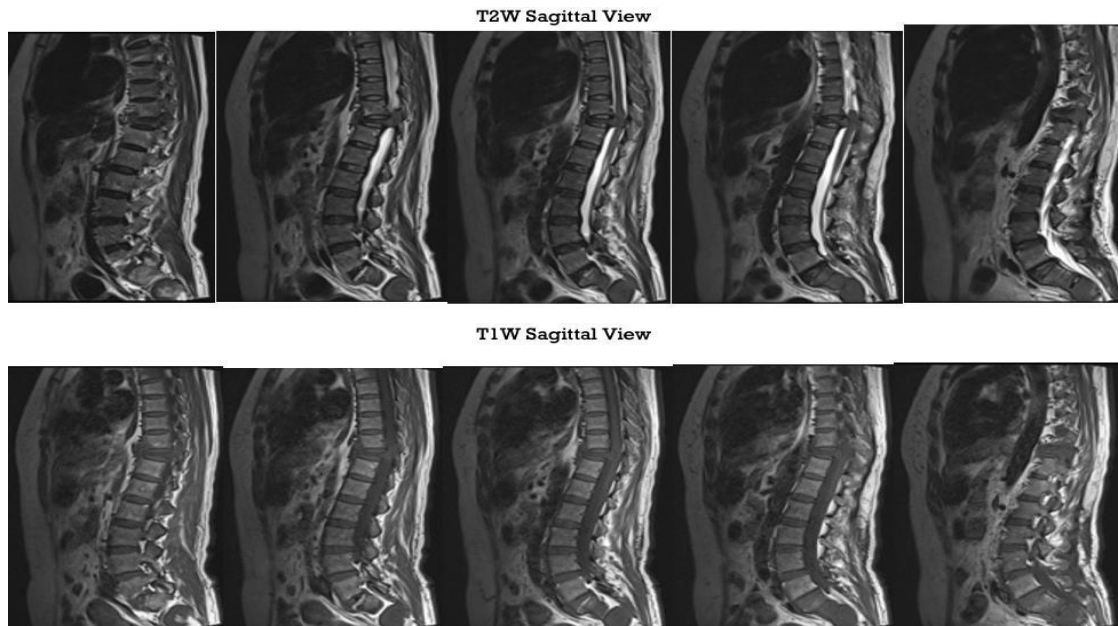


Figure 2. Magnetic resonance imaging of thoracolumbal.

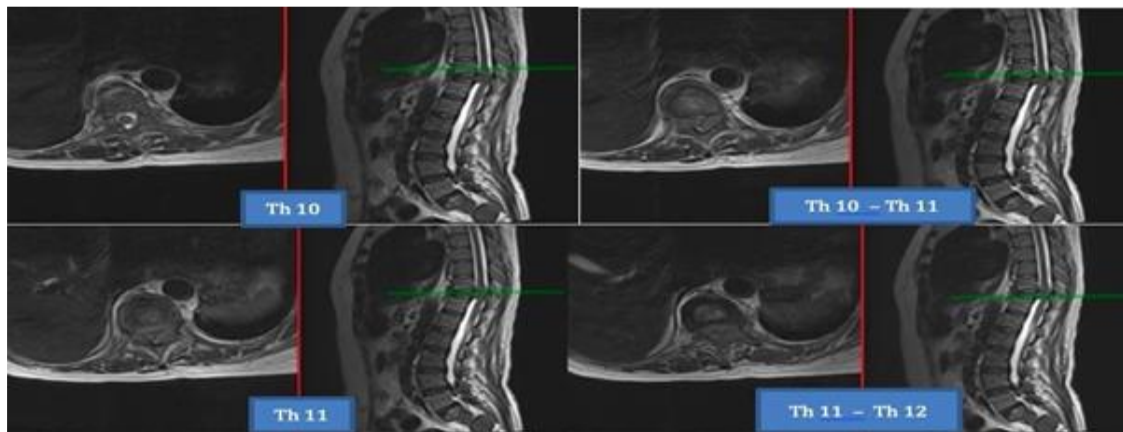


Figure 3. T2W Axial view of MRI

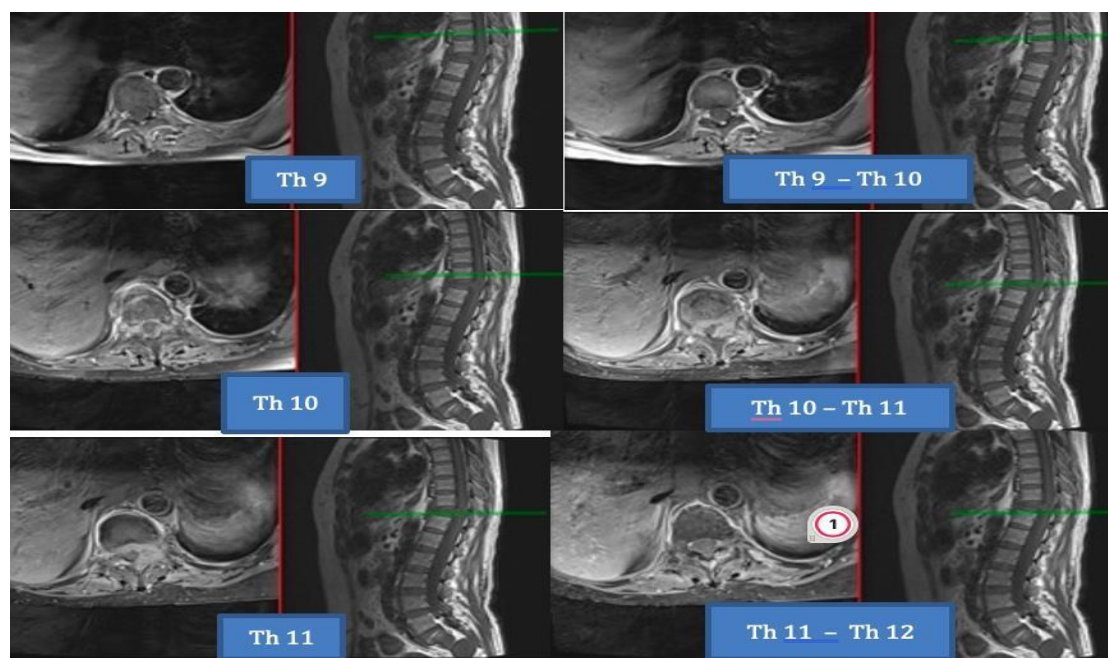


Figure 4. T1 W Axial view of MRI

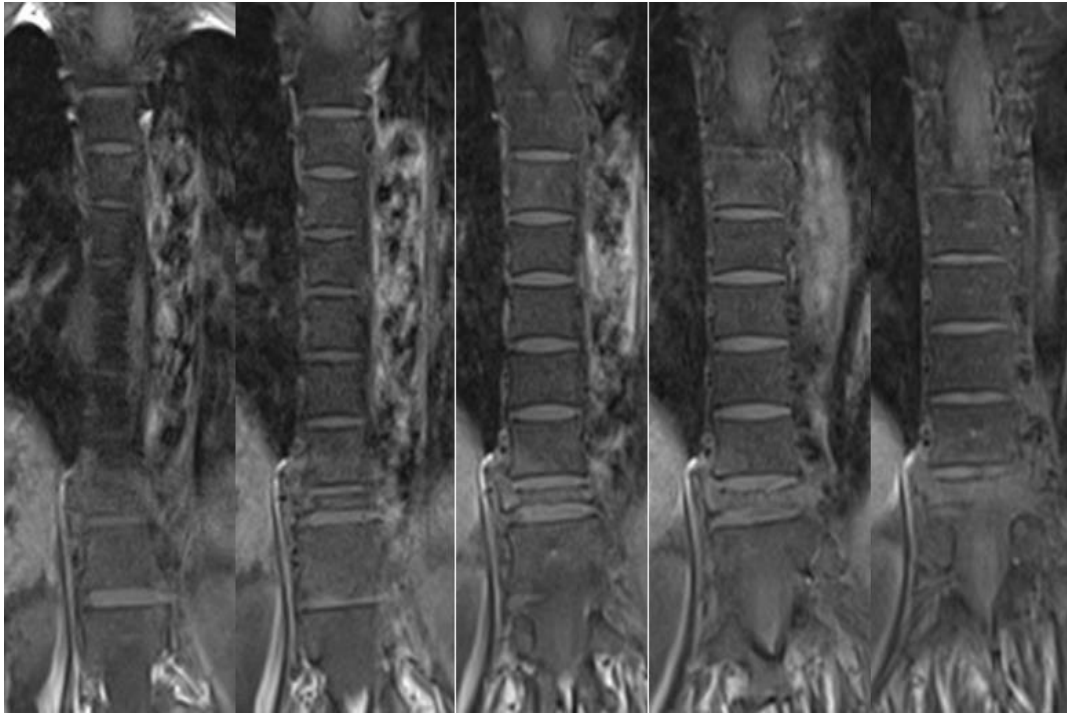


Figure 5. Coronal View of MRI

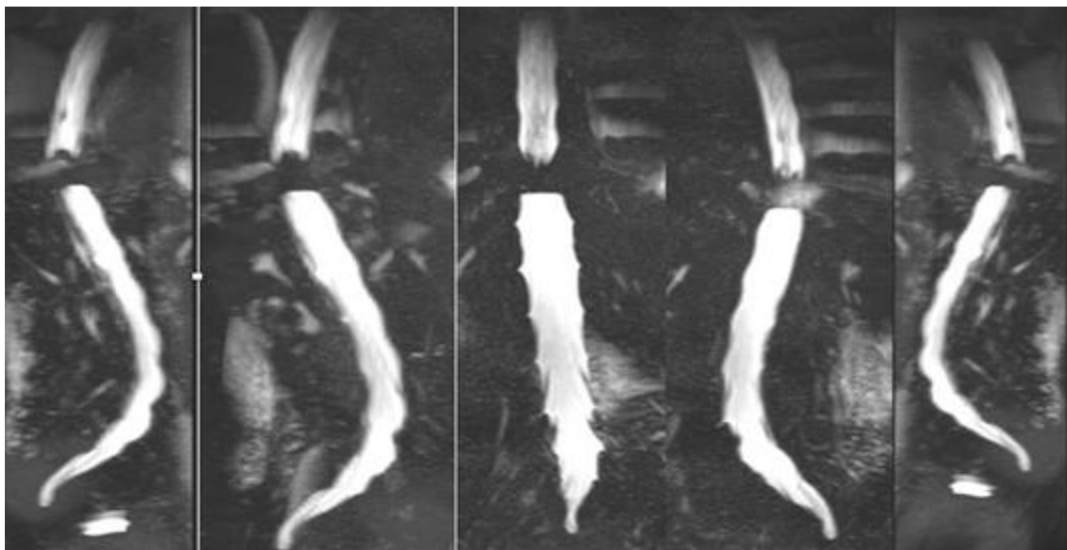


Figure 6. Myelography Examination view.

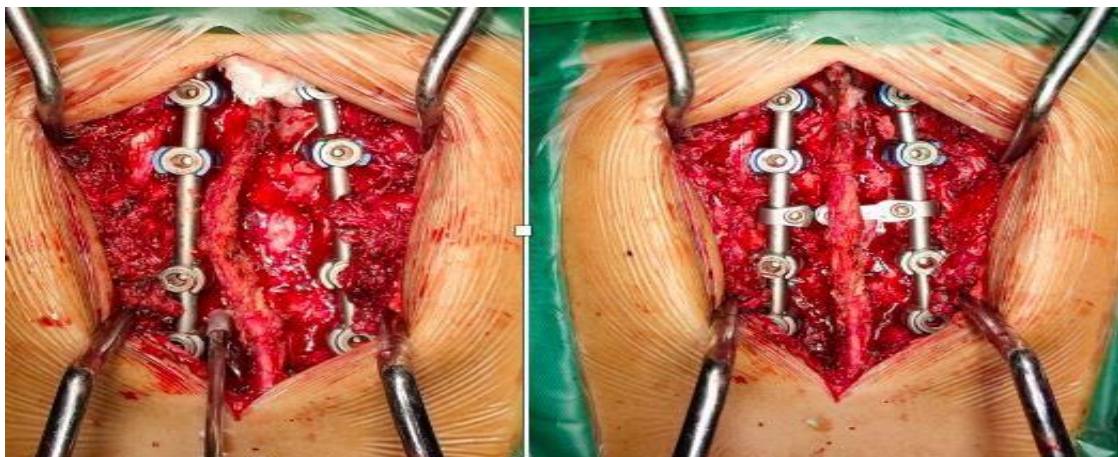


Figure 7. Intraoperatively of spine surgery at level Th10-Th11

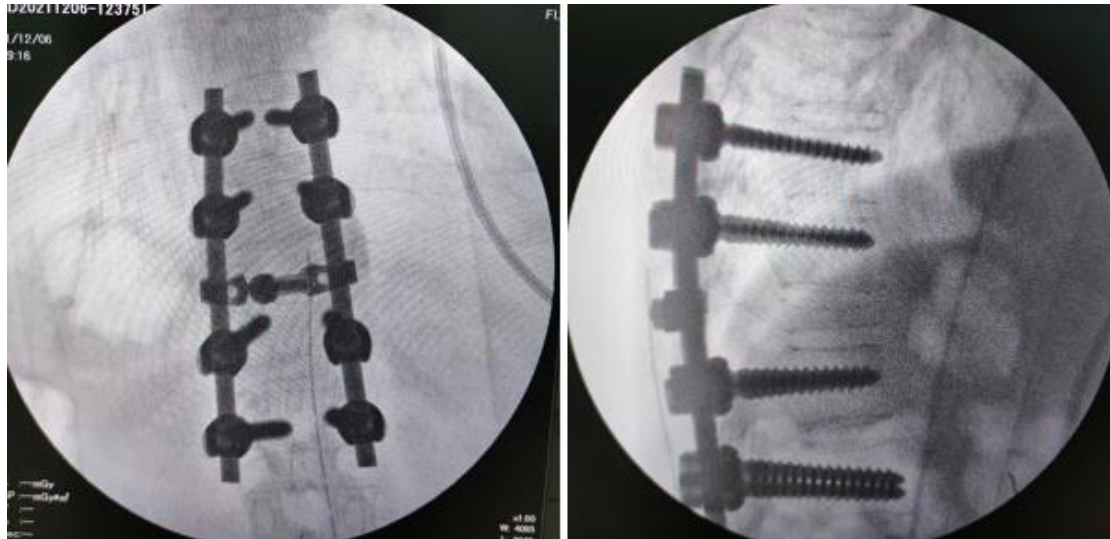


Figure 8. Intraoperatively X-Ray after decompression with stabilization fusion and biopsy.

DISCUSSION

The present study was approved by the Ethics Review Committee of The First Affiliated General Hospital of Sanglah (Bali, Indonesia), and written informed consent was obtained from the patient. Spinal 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 spinal canal arising from the postoperative field has not been reported in the previous literature. As for extra skeletal 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 a primary tumor arising in a postoperative field, which was misdiagnosed with infectious lesions and led to the 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. It 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 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 the 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 lesions 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 immunohistochemistry, 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 misdiagnose.

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 Serra¹⁴ 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.¹⁶ 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 metastatic bone disease before surgery.

CONCLUSION

In conclusion, comprehensive clinical, radiological, and laboratory investigations provide useful diagnostic information until the gold standard histopathology investigation with immunohistochemistry is established. The performance of MRI and pathologic evaluation with immunohistochemistry is particularly important for diagnosing soft tissue tumors. A follow-up chemotherapy is planned for further management of spinal osteosarcoma. Unfortunately, the patient is lost to follow-up, hence, associated with poor prognosis.

Declaration by Authors

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