



Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Primary pleomorphic liposarcoma of the spine. Case report and review of the literature

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ARTICLE INFO

Article history:

Received 22 May 2016

Received in revised form 13 June 2016

Accepted 20 June 2016

Available online 23 June 2016

Keywords:

Primary spinal tumors

Liposarcoma

Pleomorphic liposarcoma

Bone neoplasms

Total en bloc spondylectomy

Case report

ABSTRACT

INTRODUCTION: To describe a single case, the fourth ever reported, of pleomorphic liposarcoma of the spine and to undertake a review of the literature.**PRESENTATION OF CASE:** A 60 year old male patient had a bilateral lumbosciatica over a 3 month period. Imaging tests revealed a tumor mass in L1–L3 and a fracture in L2. Also, he had a mural thrombus both in the inferior vena cava and the left renal vein. The biopsy revealed a well-differentiated liposarcoma. En bloc resection of the lesion and stabilization was carried out. Due to the condition of the patient (hemodynamic instability, wound dehiscence and infection, and hypoproteinemia), a decision was made not to subject the patient to either radiation therapy or chemotherapy. The patient was subsequently found to suffer from myopathic paraparesis and a surgical wound infection. At three months, liver metastases were evident, as well as a recurrence of the lesion. A venous thrombosis that extended from the lower iliac vein to the right atrium was observed. The patient died from type I hepatorenal syndrome.**DISCUSSION:** Pleomorphic liposarcoma of the spine is a rare occurrence. En bloc resection with wide margins is the treatment of choice. The use of radiotherapy in the spine is controversial. The role that should be played by chemotherapy is still unclear, although it has been employed in treatments.**CONCLUSION:** In spite of treatment, these tumors lead to a poor prognosis, with high rates of recurrence, metastasis, and mortality.© 2016 The Author(s). Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

1. Introduction

Liposarcomas are malignant tumors found in soft tissues. They are the most common sarcomas seen in adults [1]. The pleomorphic subtype is the rarest and the most aggressive subtype [2]. There are only three cases described in which the primary form of this variant was found in the backbone [3–5]. We report a case of pleomorphic liposarcoma of the lumbar spine.

2. Presentation of case

A 61 year old male with four-month history of a bilateral lumbosciatica. A physical examination showed that there was a weakness in the lower extremities at the level of the bilateral hallucis, which was graded at 4/5. Also, there was an associated hypoesthesia at the right L2 and L3 and left L1 and L2 dermatomes. There were no perturbations in the sphincters.

An MRI of the entire spine and a CT of the lumbar spine revealed a pathological fracture in L2 and that the prevertebral tumor extended into L1 and L3 (Fig. 1). CT scans of the chest, abdomen, and pelvis confirmed no evidence of metastatic disease. CT angiography revealed a thrombus in the juxtarenal vena cava and the left renal vein (Fig. 2). A body PET-CT brought to light a tumor mass in L2 bordering L2 and L3. It also exposed a tumoral mural thrombus within the inferior vena cava. The CT-guided biopsy of the tumor mass led to an initial diagnosis of a well-differentiated liposarcoma.

The case was discussed by the hospital Tumor Committee. The large tumor mass, the chemoresistance of the well differentiated liposarcoma to adjuvant chemotherapy, and the patient's condition were taken into account. Placement of a vena cava filter followed by resection of the tumor was the course of action decided upon.

The surgery was performed in three stages. In the first stage, a right thoraco-phreno-laparotomy approach was used to perform a thrombectomy of the tumor inside the inferior vena cava and the left renal vein. After assessing the impact on the vascular wall, a vascular prosthesis (Gore-Tex) was implanted. The tumor in the right psoas was also removed. A discectomy of the L1–L2 of L3–L4 discs was performed. In the second stage of the surgery, tumor was removed at the level of the left psoas. In the immediate post-

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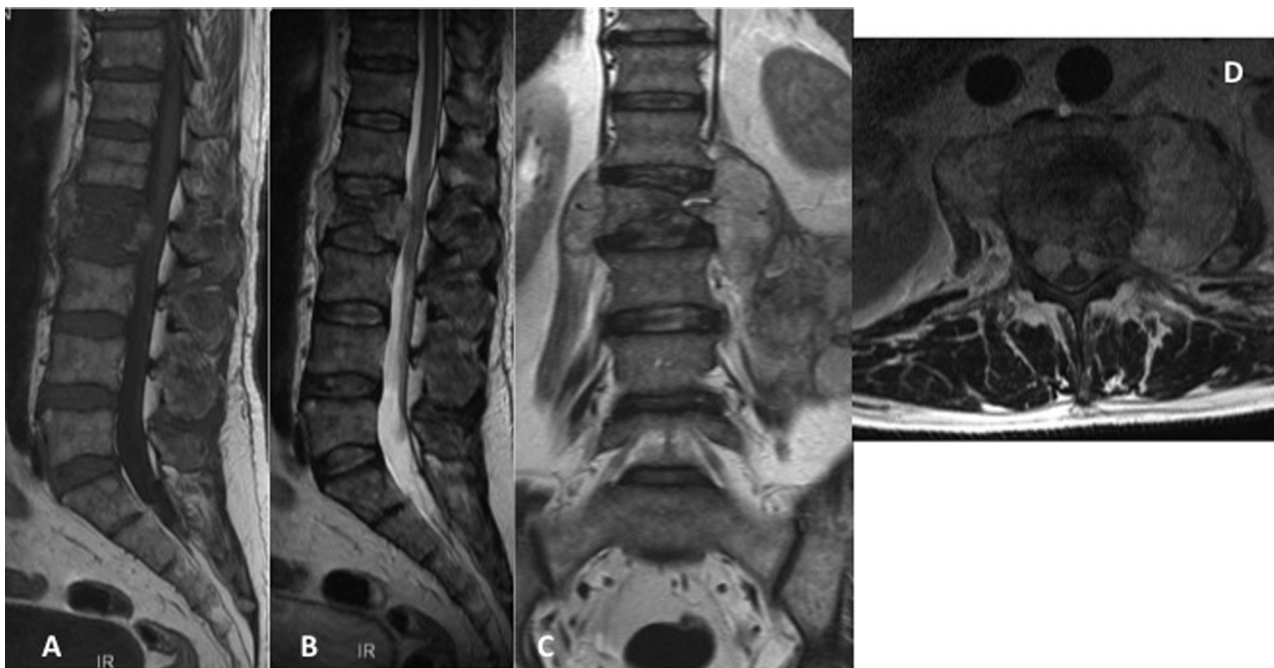


Fig. 1. MRI of the lumbar spine.

(A) A sagittal T1-weighted image which shows a hypointense signal, a fracture at the level of L2, and a prevertebral extension at the lower third of L1 and upper L3. Also seen is an anterior epidural component which contacts the L1–L2 disc and extends caudally, especially on the left side, a few millimeters from the L3 to L4 disc.

(B) A sagittal T2-weighted image showing a hyperintense signal of the bone marrow at the affected levels.

(C) A coronal T2-weighted image reveals paravertebral extension that appears predominantly on the left side of the tumor. The right paravertebral component extends from the L1 to L2 disk spaces and has a longitudinal diameter of 6.6 cm. The left paravertebral extends from the T12 to L1 disk space to L3 to L4. The rest of the impact on the left psoas appears to be due to a hematoma.

(D) An axial T2-weighted image.

operative period the patient had a pulmonary thromboembolism. Moreover, he had a retroperitoneal hematoma with a size greater than 25 cm which had to be surgically drained the next day. During the third stage of operation, an en bloc resection of tumor was performed through a posterior approach. In addition, reconstruction at T10–T12, L4, and L5 was carried out with pedicle instrumentation and at the stackable boxes of T12 to L3 with a structural allograft (Fig. 3). Tumor growth in the epidural venous plexus was monitored during this final stage of surgery.

Histological analysis tumor specimen showed that it had the features of a pleomorphic liposarcoma (Figs. 4 and 5). The maximum diameter of the tumor was 10 cm. 8 Mitoses were observed per high-powered field and tumor necrosis was observed in 50% of the total tumor area. Lymphovascular invasion was observed. The tumor impacted the right edge of the mass. Histological analysis of the mural thrombus also confirmed that its tumoral origin.

Postoperatively, the patient developed significant hemodynamic instability. This called for the transfusion of 18 red blood cell concentrates and the infusion of vasoactive drugs. The patient displayed dehiscence and surgical wounds that were infected with *Acinetobacter baumannii* and *Staphylococcus haemolyticus*. These issues were rectified in the operating room through antibiotic treatment and VAC (Vacuum Assisted Closure) therapy. The clinical outcome of these interventions was good. Regarding neurological elements, sagging was observed in the lower limbs and muscular strength was graded 0/5. Non-responsive tendon reflexes were observed in all four limbs, as were incontinence of the bladder and anal sphincters. The electromyogram was consistent with an inflammatory polymyopathy in the context of the myopathy of the critically ill patient. In light of the patient's clinical situation and its complications, adjuvant treatment options were not considered.

Over the course of treatment, the patient completely recovered muscular strength in his upper limbs while recovering only par-

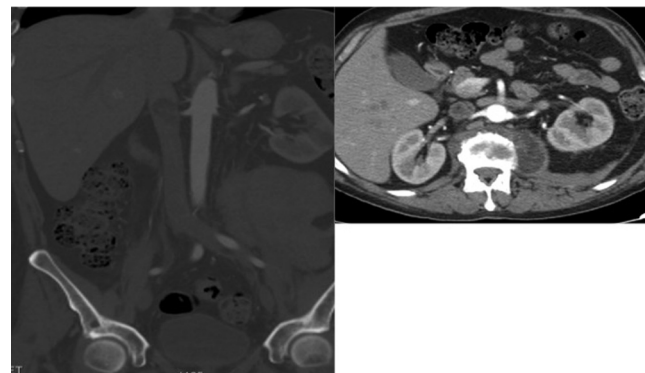


Fig. 2. A coronal and an axial CT angiogram in which thrombi are seen in the vena cava and the left renal vein.

tial strength in his lower limbs. Also, he regained control of his sphincters.

Two months after surgery, the patient progressively developed lower limb edema, kidney failure, ascites, coagulation disorder, and jaundice followed by encephalopathy. Upon revising the image study, the disease was shown to develop locally as a mass in the para-aortic area and in the area of the hematoma in the iliopsoas. There was also a single liver metastasis. Also observed was an extensive thrombosis spreading from the femoral, and external and common iliac veins to the vena cava and the right atrium. This thrombosis caused hepatic deterioration and the patient died.

3. Discussion

Liposarcoma is a malignant soft tissue tumor that originates from primitive mesenchymal cells. 20% of all adult sarcomas are

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