Int J Clin Exp Pathol 2016;9(3):4038-4042
www.ijcep.com /ISSN:1936-2625/IJCEP0021904

Case Report

Retroperitoneal liposarcoma manifested as an inguinoscrotal mass: a case report

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Received December 14, 2015; Accepted February 25, 2016; Epub March 1, 2016; Published March 15, 2016

Abstract: The inguinal canal communicates with the retroperitoneum. This creates a pathway for the extension of retroperitoneal sarcomas into the inguinoscrotal region. A 65-year-old Japanese male presented with a giant painless right inguinoscrotal mass that started growing 2 years before presentation. Computed tomography and magnetic resonance imaging revealed a giant dumbbell-shaped mass with a narrow segment in the inguinal canal connecting the retroperitoneal component with the scrotal component. En bloc surgical removal of the tumor was carried out. Histopathological analysis of the resected tumor revealed a well-circumscribed atypical lipomatous tumor in the retroperitoneum and a dedifferentiated liposarcoma in the inguinoscrotal region. In conclusion, we experienced a case of giant liposarcoma that appeared as a dumbbell-shaped tumor extending from the retroperitoneum to the scrotum through the inguinal canal. In cases involving an inguinoscrotal mass, the possibility of extension of retroperitoneal sarcoma into the inguinoscrotal region should be considered.

Keywords: Dumbbell-shaped liposarcoma, retroperitoneum, inguinoscrotal region, inguinal canal, atypical lipomatous tumor, dedifferentiated liposarcoma

Introduction

Liposarcoma is one of the most common soft tissue sarcomas found in adults. It occurs in the extremities (52%), retroperitoneum (19%) and the inguinal region (12%) [1]. The inguinoscrotal region is an uncommon location for liposarcoma [2, 3]. Here, liposarcoma manifests as a slow-growing painless mass. It can be mistaken for inguinal hernia, hydrocele, spermatocele, hematocele, epididymo-orchitis, lipoma or a tumor arising from spermatic cord or testis [4, 5]. The present report describes the case of a patient with a giant dumbbell-shaped liposarcoma extending from the retroperitoneum to the scrotum through the inguinal canal. En bloc surgical removal of the tumor was carried out via the extraperitoneal and inguinal approaches by means of a long pararectal incision that extended to the scrotum.

Case presentation

A 65-year-old Japanese male presented with a giant painless right inguinoscrotal mass that started growing 2 years before presentation. Contrast-enhanced computed tomography (CT) revealed the dumbbell shape of the mass with a retroperitoneal component contiguous with a paratesticular component across a narrow segment in the right inguinal canal (**Figure 1A** and **1B**). The retroperitoneal lesion primarily exhibited fat density with a linear non-fat component inside. The inguinoscrotal mass had enhancing heterogeneous solid tumor attenuation areas with focal calcification and a fat component inside. Magnetic resonance imaging (MRI) (**Figure 1C** and **1D**) revealed the scrotal lesion as a large heterogeneous intensity on T2-weighed images (**Figure 1D**). This scrotal lesion included a partition-like structure inside. The fat and non-fat components were contiguous and well-circumscribed. Radiologically, both testes were identified normally. The tumor was completely resected via the extraperitoneal and inguinal approaches by means of a long pararectal incision that extended to the scrotum (**Figure 2A** and **2B**). The inguinal lesion was inseparable from the spermatic cord and right testis. Therefore, right orchidectomy was also

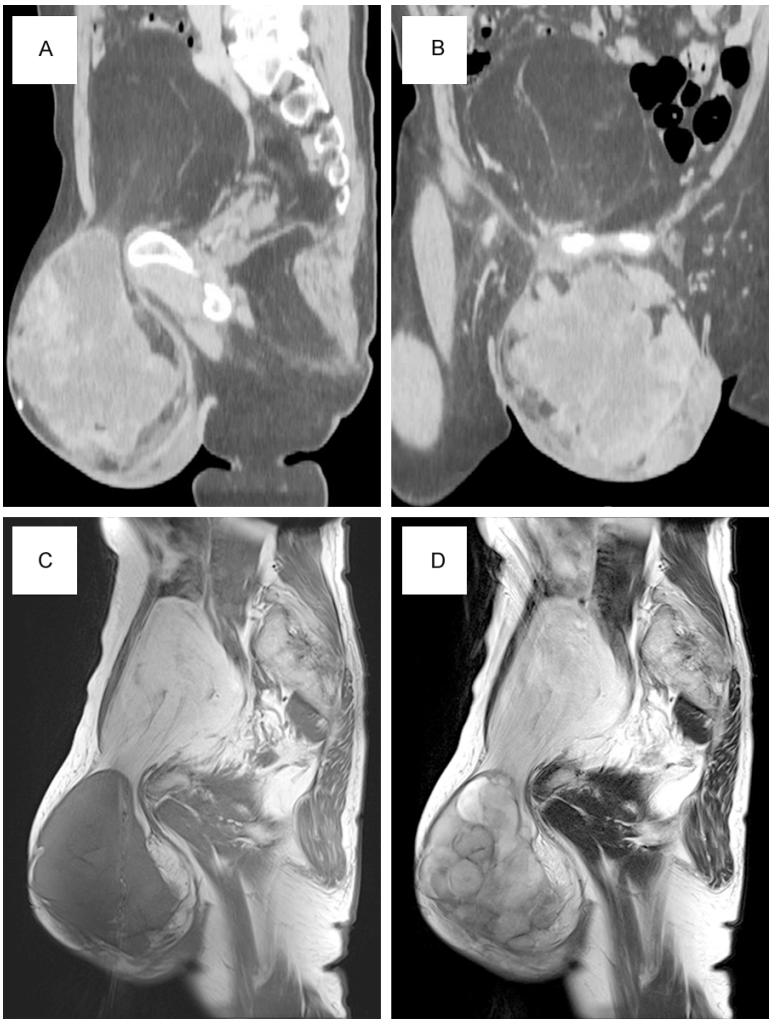

Figure 1. Computed tomography (CT) and magnetic resonance imaging (MRI) scans of the tumor. (A) Sagittal and (B) coronal contrast-enhanced CT images showing a giant dumbbell-shaped mass. The retroperitoneal lesion mostly exhibits fat density and the inguinoscrotal mass has the characteristics of an enhancing heterogeneous solid tumor. (C) T1-weighted and (D) T2-weighted-sagittalMR images revealing a separated biphasic pattern involving different components. The high signal intensity on T2-weighted images suggested a mucus component.

carried out. The right inguinal ligament was preserved. The weight of the resected specimen was 2.8 kg. Histopathological analysis of the retroperitoneal lesion revealed a mixture of normal-appearing adipocytes intermixed with atypical adipocytes. It was diagnosed as an atypical lipomatous tumor (**Figure 3A**). The structure of the spermatic cord was destroyed and unclear. The well-differentiated and poorly differentiated components showed a steep transition in the region that appeared to be the spermatic cord (**Figure 3B**). The scrotal lesion exhibited an extensive poorly differentiated component that involved necrotic tissue (**Figure 3C**). Spindle-shaped atypical cells had proliferated and exhibited myxoma-like stroma (**Figure 3D**). In addition, a well-differentiated component showing nodular hyperplasia was also observed; immunohistochemical evaluation revealed that cells were positive for murine double-minute type 2 (MDM2) and cyclin-dependent kinase 4 (CDK4) (**Figure 3E** and **3F**). The inguinoscrotal lesion was diagnosed as a dedifferentiated liposarcoma. The postoperative course was uneventful and the patient was discharged on the 10th postoperative day. No recurrence was observed for 6 months after surgery.

Discussion

Liposarcomas are malignant mesenchymal tumors of adipose origin. Among the retroperitoneal sarcomas, liposarcoma is the most frequent histological type comprising 41% of these tumors [6]. As was the case in the present patient, this tumor usually occurs in individuals aged 40–60 years. Areas of predilection for liposarcomas are the extremities, retroperitoneum and inguinal region [2, 3, 7]. According to the 2013 World Health Organization's classification, liposarcomas are histologically classified into four subtypes (atypical lipomatous tumor/well-differentiated liposarcoma, dedifferentiated liposarcoma, myxoid liposarcoma and pleomorphic liposarcoma) based on their natural history, morphological features and cytogenetic alterations. Pleomorphic and dedifferentiated subtypes are regarded as high-grade; whilst atypical lipomatous tumors and myxoidliposarcomas are regarded as low-grade [8, 9]. The classification of liposarcoma is quite important, because the size and histological subtype

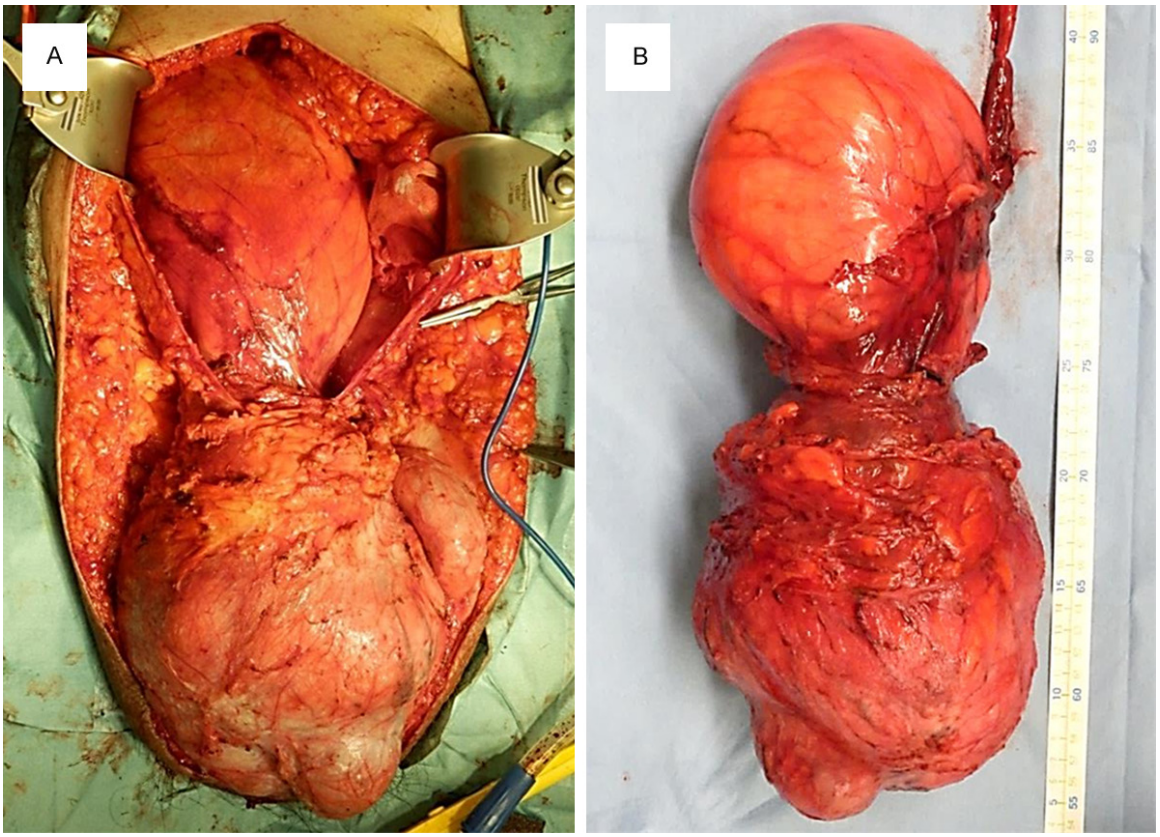

Figure 2. Photographs showing (A) intraoperative findings and (B) the gross appearance of the resected tumor.

determines the treatment and prognosis. Primarily, liposarcomas have a tendency to spread by local extension. Generally, hematogenous and lymphatic spread is a late event associated with high-grade tumors. Atypical lipomatous tumors and myxoid liposarcoma may recur locally and their metastatic potential is low. In contrast, pleomorphic and dedifferentiated subtypes have metastatic potential, which may reduce the survival rate [8]. In our present case, the patient had no local recurrence or metastatic disease at6 months after the surgery.

In the evaluation of liposarcoma, radiological imaging including CT and MRI is useful. CT is helpful as a preoperative diagnostic tool in liposarcoma because it can be used to identify the characteristic attenuation of fat within the retroperitoneal and inguinal lesion. Attenuation of liposarcoma on CT images, especially the amount of fat in the tumor, reflects its histological subtype [7, 10]. As malignancy progresses, liposarcomas contain less fat and show attenuation similar to that of muscle [2, 7]. However, there have only been a few case reports regarding radiologic findings for retroperitoneal dedifferentiated liposarcoma. Hong et al. [2] evaluated the spectrum of radiological appearance of retroperitoneal dedifferentiated liposarcoma in combination with clinicopathological features. They retrospectively studied the radiological images and clinical histories of 15 patients with histologically verified retroperitoneal dedifferentiated liposarcoma. They classified the appearance of the tumor as follows: Category I, a non-fatty component within a predominantly fatty mass (n=5); Category II, a focal fatty component within a largely non-fatty mass (n=6); Category III, a well-defined fatty mass and well-defined non-fatty mass (n=1); and Category IV, two masses with a predominantly nonfatty component (n=3). In our present case, contrast-enhanced CT revealed the differences in fat attenuation between retroperitoneal and inguinoscrotal lesions. It clearly reflected histological subtypes. Moreover, MRI revealed a separate biphasic pattern for dedifferentiated liposarcoma and atypical lipomatous tumors.

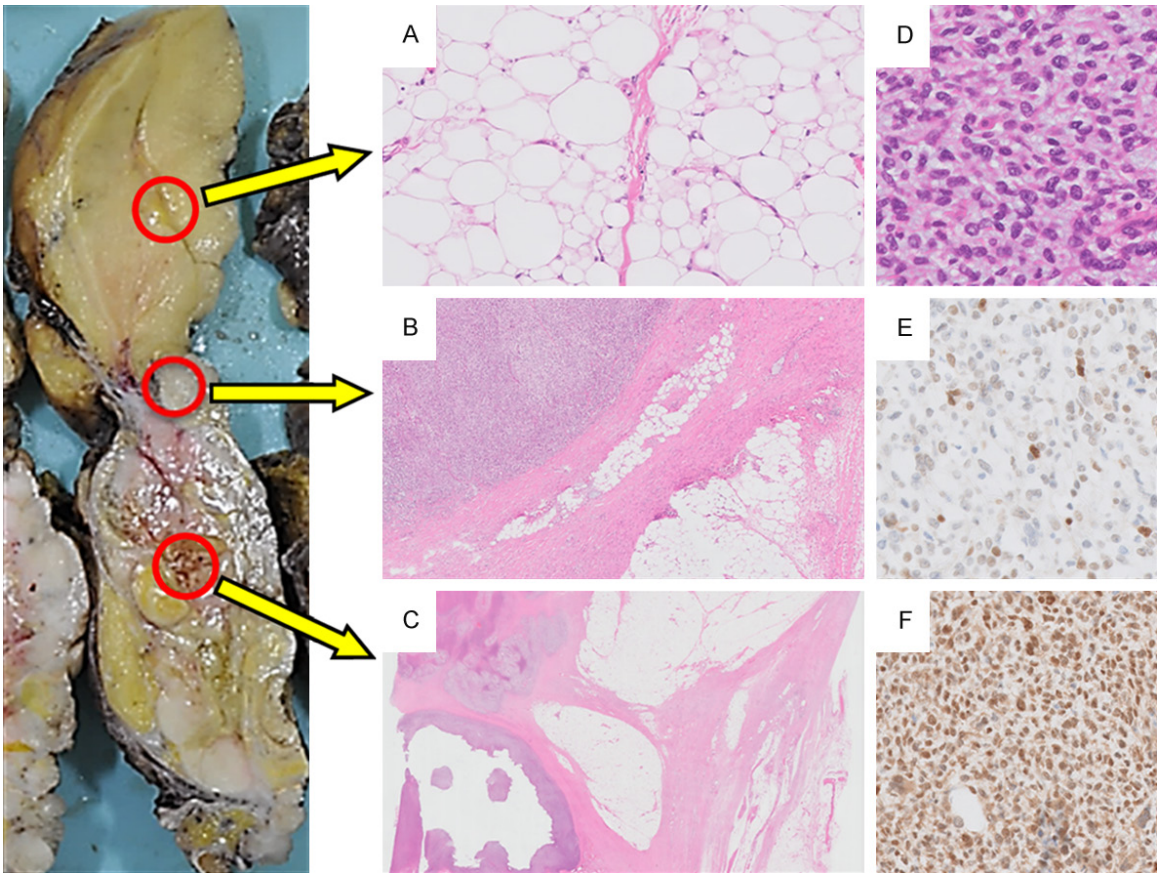

Figure 3. Histopathological findings from the resected tumor. Photomicrographs of tissue sections from (A) the retroperitoneal lesion, (B) the spermatic cord lesion, and (C) the scrotal lesion stained with hematoxylin and eosin. (D) Positive immunohistochemical staining of tissue sections from the scrotal lesion with (E) MDM2 and (F) CDK4.

Our case falls under Category III (a well-defined fatty mass and well-defined non-fatty mass). Thus, our findings also supported the opinion that CT and MRI are useful tools in determining pathological features at diagnosis.

The prognosis of patients with liposarcoma remains poor. Even after complete resection, the 5-year survival rate of patients with retroperitoneal atypical lipomatous tumors is 83%, while it is 20% for the dedifferentiated tumor subtype [8]. The main cause of mortality in retroperitoneal liposarcoma has been reported to be local recurrence [11]. Therefore, to achieve complete resection for local control, combined resection of the surrounding organs may be unavoidable. For the detection of recurrence, a CT scan every 3 months for the first 2 years, every 6 months from 2-5 years, and annually thereafter is generally recommended [9].

In conclusion, we reported a case of giant dumbbell-shaped liposarcoma extending from the retroperitoneum to the scrotum through the inguinal canal. CT and MRI scans are helpful as a preoperative diagnostic tool in liposarcoma by identifying tumor characteristics. In cases involving an inguinoscrotal mass, the possibility of retroperitoneal sarcoma extension into the inguinoscrotal region should be considered.

Acknowledgements

This study was not supported financially by any private funding agency.

Disclosure of conflict of interest

None.

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