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Case Report

Incidentally discovered well-differentiated retroperitoneal liposarcoma with inguinal canal herniation: report of 2 cases

Valeria Fiaschetti MD, PhD^a, Francesca Castellani MD^{a,*}, Giusy Croce MD^a,
 Mariateresa Mondillo MD^a, Andrea Amico MD^a, Adriano De Majo MD, PhD^b,
 Valentina Girardi MD^a, Roberta Di Trapano MD^a, Roberto Floris MD, PhD^a

^a Department of Radiology, Tor Vergata Polyclinic, Viale Oxford 81, 00100 Rome, Italy

^b Department of General Surgery, Tor Vergata Polyclinic, Rome, Italy

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ABSTRACT

Well-differentiated retroperitoneal liposarcomas are slow growing and low-grade tumors, reaching usually huge size before being symptomatic and so diagnosed, therefore with increase of the surgical risk and of the probability of dedifferentiation. Inguinal location of these tumors is unusual and rarely diagnosed.

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In this study, we report 2 cases of incidental finding, during emergency room access, of retroperitoneal liposarcoma herniated in the left inguinal canal, documented with computed tomography with and without contrast medium administration. The final histological examination was respectively “sclerosant” and “lipoma-like” well-differentiated liposarcoma.

Imaging is currently the first step for a correct diagnosis and subsequent treatment, identifying any components of the dedifferentiation of the tumor and allowing an appropriate pre-operative evaluation that appear to be the main prognostic factor.

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* Corresponding author.

E-mail address: francescastellani@libero.it (F. Castellani).

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Case report

Case 1

A 64-year-old male patient reached our emergency room with right renal colic pain symptoms worsening in the last hours. A baseline computed tomography (CT) confirmed the presence of a right ureteral stone close to the ureteral ostium with ectasia of the ipsilateral ureter. Moreover, CT showed the presence of a retroperitoneal mass in the left pelvis (Fig. 1).

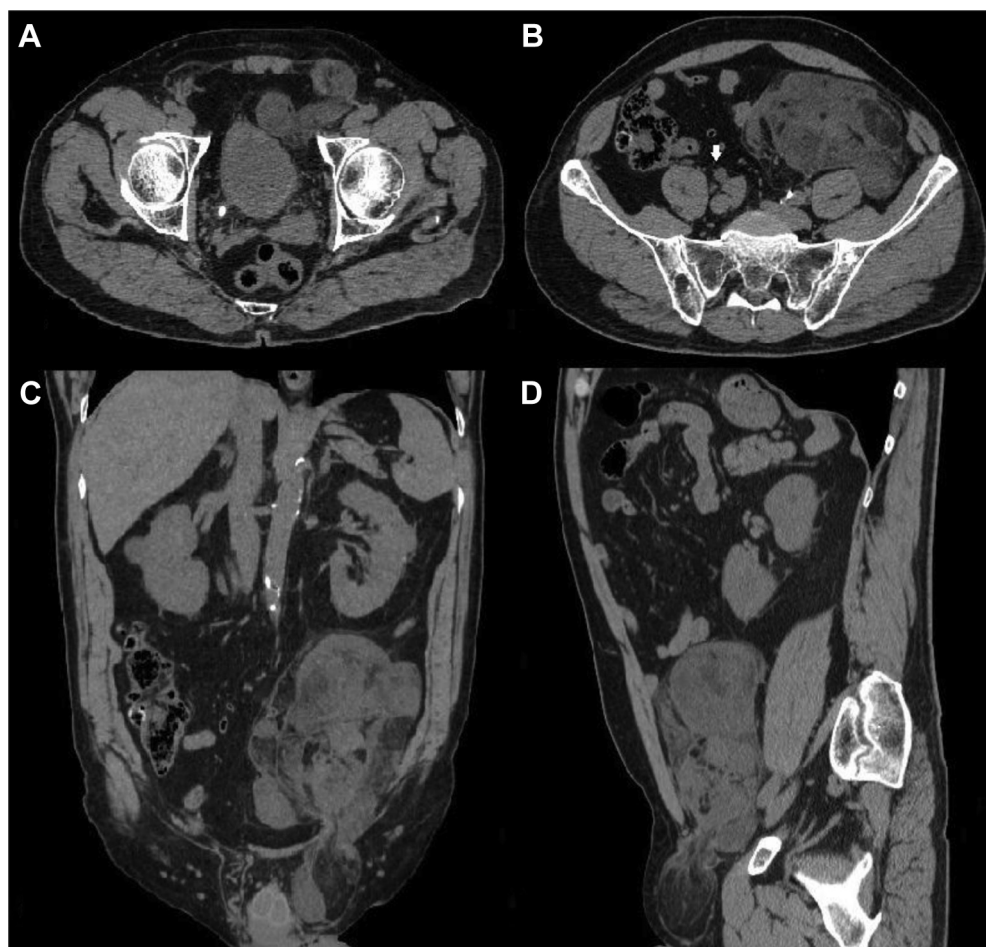


Fig. 1 – Baseline axial CT shows right ureteral stone (A) with distention of the ipsilateral ureter (B, white arrow). CT also shows the presence of a partially adipose retroperitoneal mass located in the left iliac region. Coronal (C) and sagittal (D) CT showing the retroperitoneal mass herniated through the left inguinal canal to the ipsilateral scrotal region. CT, computed tomography.

Supplemental CT investigation, after contrast medium administration (Fig. 2), confirmed the presence of an encapsulated retroperitoneal mass, measuring $13 \times 21 \times 19$ cm, with inhomogeneous density, septa, and nodular foci (53 mm and 40 mm approximately) of early and intense contrast enhancement, and some areas of fat tissue content. This finding extended from the left iliac region through the ipsilateral inguinal canal to the scrotal region where it was located a further solid nodule of 32 mm. The bladder, the bowel loops, the colon, the colonic sigma, and rectum appeared displaced medially and upwards without signs of infiltration.

The lesion densitometric characteristics supported the hypothesis of retroperitoneal liposarcoma. The abdominal study did not reveal swellings lymph nodes or images related to secondary tumors.

The patient underwent surgical removal of the mass. The definitive histological diagnosis was sclerosing well-differentiated retroperitoneal liposarcoma with disease-free margins.

Case 2

A 67-year-old male patient arrived at our emergency room department for severe pain in the left thigh. He had a history of left inguinal hernia surgery. The physical examination detected a bulge in the left groin area, which was not manually reducible at the maneuver of Taxis.

Ultrasound examination showed solid tissue, with mixed echostructure and intense vascularization in the left groin region.

The patient underwent CT before and after administration of contrast medium that showed the presence of a retroperitoneal mainly adipose mass, with dimension of $20 \times 24 \times 21$ cm and extension from the mesogastric and left lumbar to the left iliac region with herniation in the ipsilateral inguinal canal (Fig. 3).

This lesion was well encapsulated with parenchymal inhomogeneous structure, mainly adipose, within thin septa and some hyperdense nodules, the largest about 30 mm wide, characterized by early and intense contrast

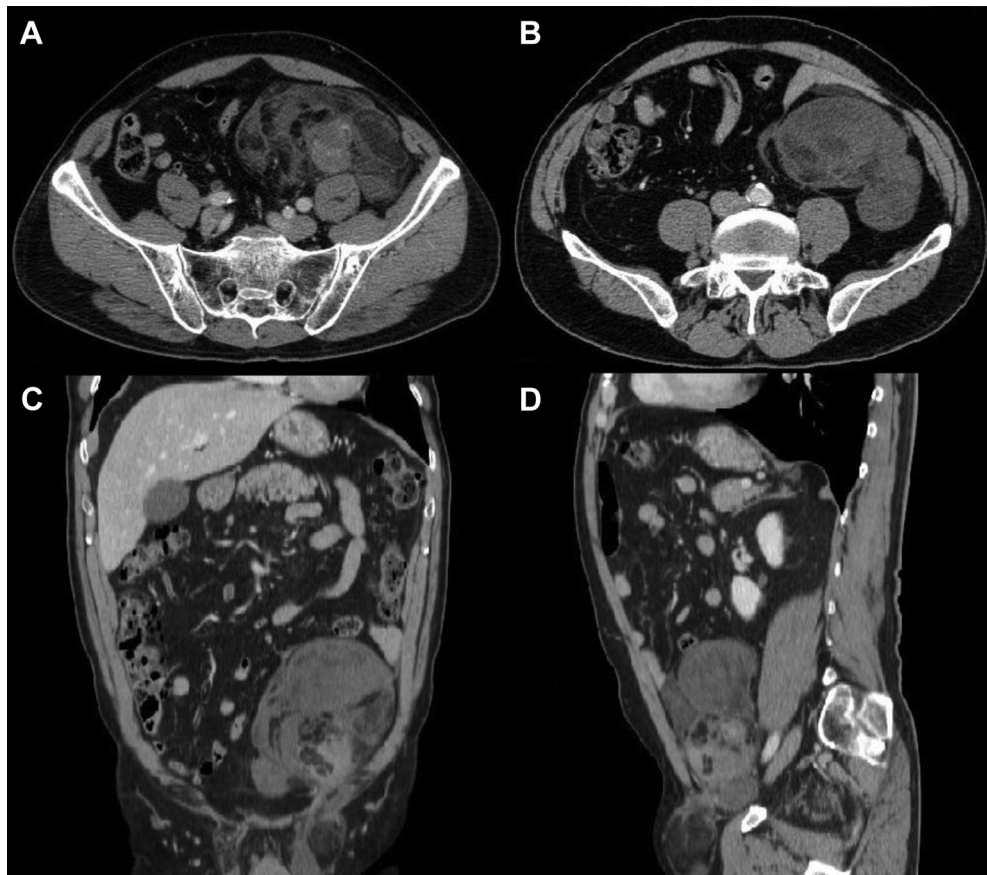


Fig. 2 – Axial (A and B), coronal (C), and sagittal (D) CT images after contrast medium administration, showing soft-tissue component and foci and septa of contrast enhancement. Abdominal evaluation did not reveal other organs abnormalities. CT, computed tomography.

enhancement. The mass caused compression and dislocation of the following structures: transverse colon, descending colon, bowel loops, abdominal aorta, left iliac vessels,

bladder and ipsilateral ureter without significant dilatation of the renal pelvis, or abdominal organs infiltration signs (Fig. 4).

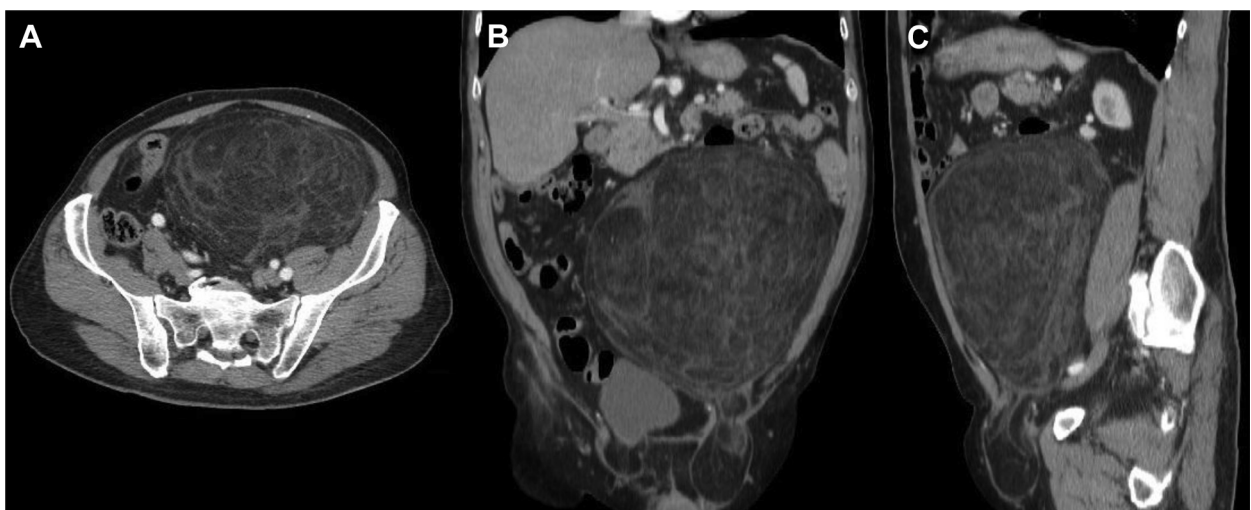


Fig. 3 – Contrast-enhanced CT (A-C) shows the presence of a giant retroperitoneal capsulated, mainly adipose, mass with thin septa and some nodular foci with early contrast enhancement. This lesion presents herniation in the left inguinal canal. CT, computed tomography.

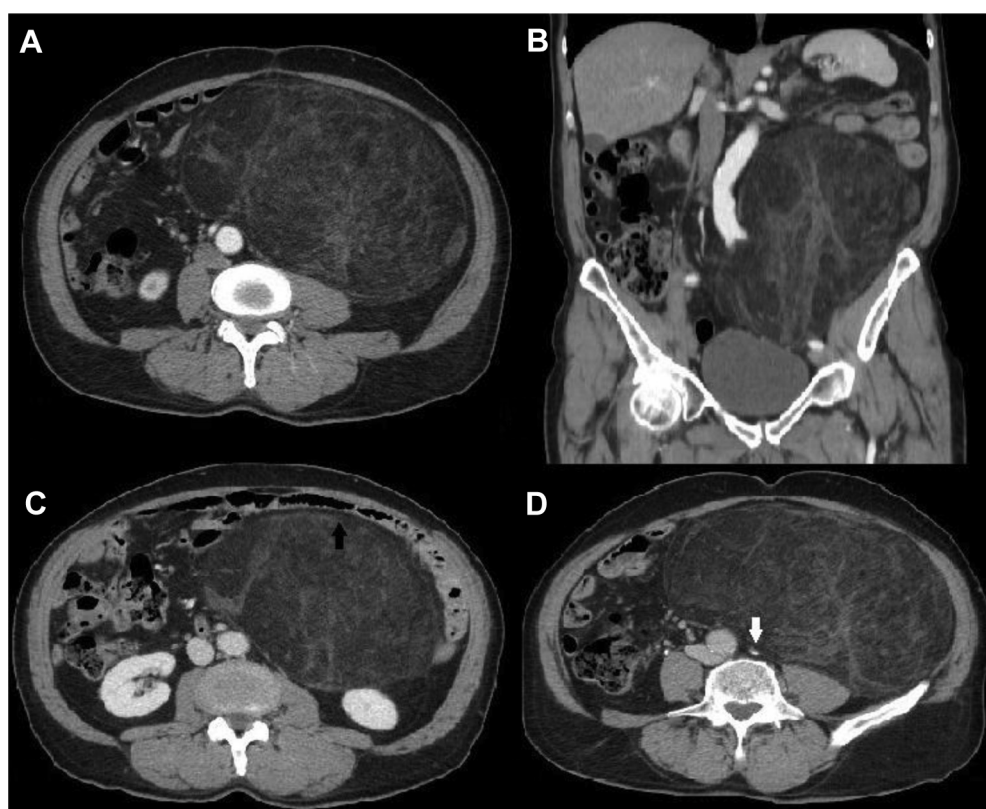


Fig. 4 – Axial and coronal contrast-enhanced CT showing displacement of the aorta (A and B), of the colon (C, black arrow), of the sigmoid colon, of the bladder and of the left ureter (D, white arrow) which results regularly pervious. CT, computed tomography.

The patient underwent surgery (Fig. 5), and the definitive histological diagnosis was “lipoma-like” well-differentiated retroperitoneal liposarcoma with disease-free margins.

Discussion

Inguinal liposarcomas are rare tumors, resulting in less than 6.6% of all retroperitoneal liposarcomas [1]. In this study, we reported 2 cases of retroperitoneal liposarcoma herniated in the left inguinal canal. Incidental finding of an abdominal and/or groin mass needs an accurate differential diagnosis with benign and malignant diseases of the retroperitoneal space and of the inguinal region, such as lipoma, teratoma, angiomyolipoma, myelolipoma, malignant fibrous histiocytoma, leiomyosarcoma, lymphoma, and retroperitoneal hematoma. In both patients, we had a large abdominal mass and we hypothesized the retroperitoneal fat tissue to be the origin of the tumor, as confirmed by histological evaluation.

Liposarcoma is the most common type of soft-tissue sarcoma (STS) in adults, comprising 24% of extremity STS and 45% of retroperitoneal STS [2]. Incidence of liposarcoma increases with the age with most of the cases occurring between 50 and 60 years and it is more frequent in males [3].

The etiology in most cases is unclear. However, numerous studies have associated the different subtypes of liposarcomas with cytogenetic abnormalities. Liposarcomas can develop in any location of the body although they are generally located in

the head, neck, trunk, mediastinum, upper and lower extremities, gastrointestinal tract, and retroperitoneum [4].

Retroperitoneal liposarcoma often appears as an asymptomatic abdominal mass, or patients may present symptoms caused by the effect of the growing mass on adjacent structures [3]. The dimensions and weight of liposarcomas are variable, and they may get to large in the retroperitoneal space [5].

According to the 2002 World Health Organization histological classification of tumors, liposarcoma can be divided into 5 categories: atypical lipomatous tumour/well-differentiated; dedifferentiated; myxoid; pleomorphic; and mixed-type liposarcoma.

Well-differentiated liposarcomas are subdivided into 4 histological subtypes: lipoma-like, sclerosing, inflammatory type, and spindle-cell liposarcoma [6].

Mortality is highly variable as well as the recurrence of the disease and is closely related to histological type, only complete excision represents the gold standard of treatment [7].

The aim of the 2 cases here reported is to show the role of imaging in retroperitoneal liposarcoma diagnosis, staging, and surgical planning.

For liposarcomas, surgical removal is mandatory, but complete surgical resection may be difficult and recurrence is common. The main survival prognostic factors are the histologic subtype and disease-free margin of resection [8].

Radiologists have an important role for the preoperative diagnosis and also in correctly assessing the extent of the tumor. An accurate identification of the extent of the tumor is



Fig. 5 – Surgical images show resection of the mass and the surgical specimen.

crucial because incomplete resection frequently leads to local recurrence with adverse outcome on the prognosis [9].

The imaging appearances of liposarcomas change depending on the histological subtype. On ultrasound, liposarcoma appears as a heterogeneous, multilobulated soft-tissue mass. The presence of hyperechoic foci indicates that the mass is lipomatous in nature, but ultrasonography is not enough for a full assessment of the tumor.

On CT, well-differentiated liposarcoma appears as a predominantly adipose soft-tissue mass with nonlipomatous components. Nonlipomatous features include septa (often >2 mm) and/or small foci of nodular or globular nonadipose tissue. In addition, calcifications may be present within the lesion [10]. Dedifferentiated liposarcomas can arise within the context of well-differentiated liposarcomas indeed they often have similar radiological features. However, foci of non-lipomatous tissue >2 cm in size can indicate that the lesion is a dedifferentiated liposarcoma, although this diagnosis needs to be verified histologically [2].

In our cases, CT was compatible with well-differentiated liposarcoma, because of the presence of both lipomatous and nonlipomatous components including septa and small nodular foci, especially in the second case report, in which the tumor has a predominant lipomatous component.

It is essential to identify CT signs of the histological subtype of liposarcoma because this is the most important prognostic factor. Outcomes vary widely depending on the liposarcoma subtype: well-differentiated liposarcoma has the best prognosis with 5-year survival rates of 90% or higher, whereas pleomorphic and dedifferentiated liposarcomas have 5-year survival rates reported to be as low as 30% and have high metastatic potential and recurrent locally [10,11].

The only current treatment option known to prolong survival in patients with retroperitoneal liposarcomas is wide surgical resection. Although the efficiency of preoperative and postoperative radiotherapy and chemotherapy is still a controversial issue, wide surgical resection for treatment of retroperitoneal tumors remains the gold standard procedure [12].

In our experience, imaging resulted to be basic not only for the diagnosis but also for a proper evaluation of the extent of the tumor, especially the identification of the herniated pathological tissue in the left inguinal canal, allowed an accurate surgical planning. CT locoregional staging permitted a complete tumor resection that is a very important positive prognostic factor as it greatly reduces the rate of local recurrence.

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