Radical resection for pleomorphic liposarcoma of preperitoneal fat origin: a case report

Yasumasa Yoshioka, Tadao Tokoro, Yusuke Makutani, Ryotaro Ogawa, Masayoshi Iwamoto, Toshiaki Wada, Koji Daito, Kazuki Ueda, Junichiro Kawamura

Division of Colorectal Surgery, Department of Surgery, Kindai University Faculty of Medicine, Osaka, Japan

Abstract

Introduction: **Liposarcomas are frequently occurring tumors that account for 20% of malignant soft tissue tumors.[1, 2] They frequently occur in the thigh, popliteal region, buttocks, retroperitoneum, and other areas,[3, 4] with very rare cases of tumors originating from the preperitoneal fat. The World Health Organization (WHO) has classified liposarcoma by their histological types as well-differentiated, myxoid, round cell, pleomorphic, and dedifferentiated.[4] The pleomorphic type is relatively rare and is associated with poor outcomes.[5] Here, we report a very rare case of a pleomorphic liposarcoma originating from the preperitoneal fat.**

Presentation of Case: **A 48-year-old man was admitted to our hospital with a left abdominal mass. Contrast-enhanced computed tomography of the abdomen revealed a mosaically enhanced tumor, 7 cm in diameter, between the dorsal side of the left rectus abdominis muscle and the ventral side of the inferior epigastric artery and vein. The tumor was surgically removed. Histopathological examination revealed a pleomorphic liposarcoma originating from the preperitoneal fat.**

Discussion: **Liposarcomas frequently occur in the thigh, popliteal region, buttocks region, retroperitoneum, and other areas. Reports of pleomorphic liposarcomas originating from the preperitoneal fat are very rare. In this case, the patient remains alive without tumor recurrence 5 years after the surgery because of the complete tumor resection.** ***Conclusion:*** **For tumors originating from the preperitoneal fat, surgical removal needs to be considered if there is a possibility of the lesion being a liposarcoma, as in this case.**

Key words: pleomorphic liposarcoma, peritoneal fat, computed tomography imaging, positron emission tomography imaging, magnetic resonance imaging, inferior epigastric arteries and veins

Presentation of Case

A 48-year-old man became aware of a tumor in his left abdomen and recognized that it had been increasing in size over a three-week period. Therefore, he visited his local doctor and was referred to our department for a detailed examination and treatment. An elastic, hard mass, approximately 7 cm in diameter, was found in his left abdomen. Abdominal computed tomography (CT) showed a well-defined tumor on the dorsal side of the patient's left rectus abdominis muscle; the inferior epigastric arteries and veins ran along the dorsal side of the tumor (Figure 1). A positron emission tomography (PET)-CT examination revealed a maximum standardized uptake value (SUV) of 9.95 in the same area (Figure 1). Abdominal magnetic resonance imaging (MRI) revealed a lobulated tumor with low signal intensity in the T1-

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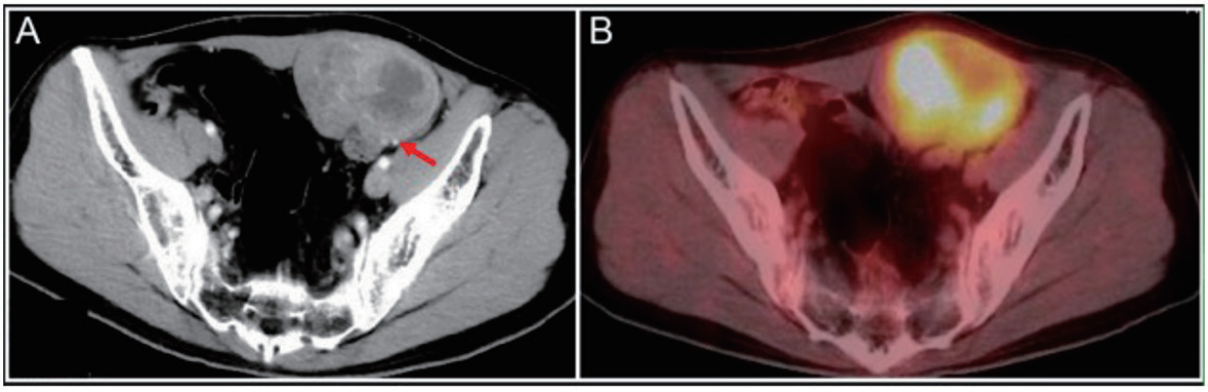

Figure 1 Initial abdominal imaging.
A: Abdominopelvic contrast-enhanced computed tomography showing a well-circumscribed, mosaic soft tissue mass beneath the left rectus abdominis muscle. The inferior epigastric arteries and veins (red arrow) travel along the dorsal side of the tumor.
B: [^{18}F] Fluorodeoxyglucose-positron emission tomography imaging showing a maximum standardized uptake value (SUV) of 9.95 in the region of the mass.

weighted scan and an inhomogeneous high-intensity signal in the T2-weighted scans. Due to the presence of preperitoneal fat tissue just beneath the tumor, the tumor was considered to have originated from the preperitoneal fat (Figure 2).

To resect the tumor, surgery was initiated through a midline incision in the lower abdomen. The observed tumor was fist-sized, protruded into the abdominal cavity, and was covered by the peritoneum; the inferior epigastric arteries and veins ran along the dorsal side of the tumor. To prevent direct exposure to the tumor wall, a complete resection was performed to remove the tumor along with the covering sections of peritoneum, posterior sheath of the rectus abdominis muscle, and the abdominal muscle. The excised specimen measured 10 × 7 × 5 cm, weighed 240 g, and had a cut surface that was yellowish white, solid, and segmented (Figure 3).

A histopathological examination revealed that the tumor tissue had a myxoid matrix and a mixture of cells with highly pleomorphic, atypical nuclei. There were areas where polynuclear cells proliferated, as well as areas with a dense population of cells with spindle-shaped, atypical nuclei. Additionally, atypical cells (lipoblasts) with vacuolated cytoplasm were found in the myxoid matrix. The tumor cells consisted of lipoblast-like tumor cells and tumor-like proliferation of cells showing pleomorphism, leading to the diagnosis of pleomorphic liposarcoma. Immunohistochemistry revealed that the cells were negative for S-100, MDM2, SMA, Desmin, CD34, and HHF35; there were a few cells that were slightly positive for CDK4. The tumor was therefore confirmed to be a pleomorphic liposarcoma, originating from the preperitoneal fat by two pathologists. The excision

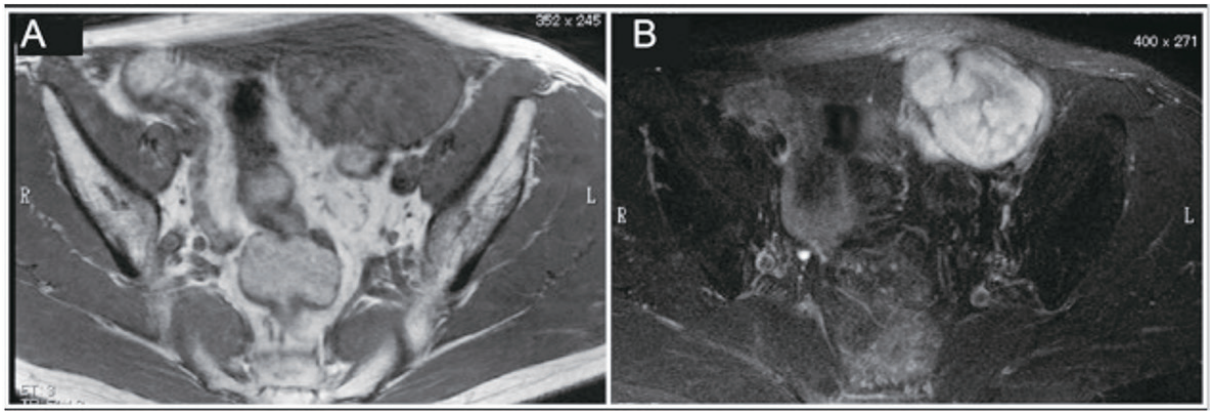

Figure 2 Abdominal magnetic resonance imaging.
A: T1-weighted image showing low signal intensity associated with a lobulated mass.
B: T2-weighted image showing inhomogeneous high signal intensity.

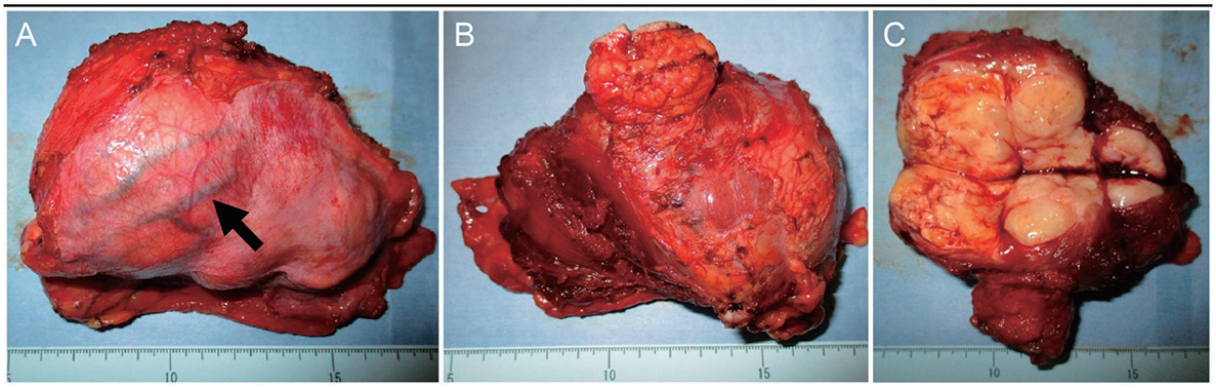

Figure 3 **Excised tumor (measuring 10 × 7 × 5 cm and weighing 240 g).**
A: The dorsal side of the tumor is shown and is covered by peritoneum; the abdominal wall arteries and veins can be seen (black arrow).
B: The ventral side of the tumor is covered by the rectis abdominus muscle; the surgical margins are grossly negative.
C: The cut surface of the tumor is yellowish white and demonstrates a lobular structure.

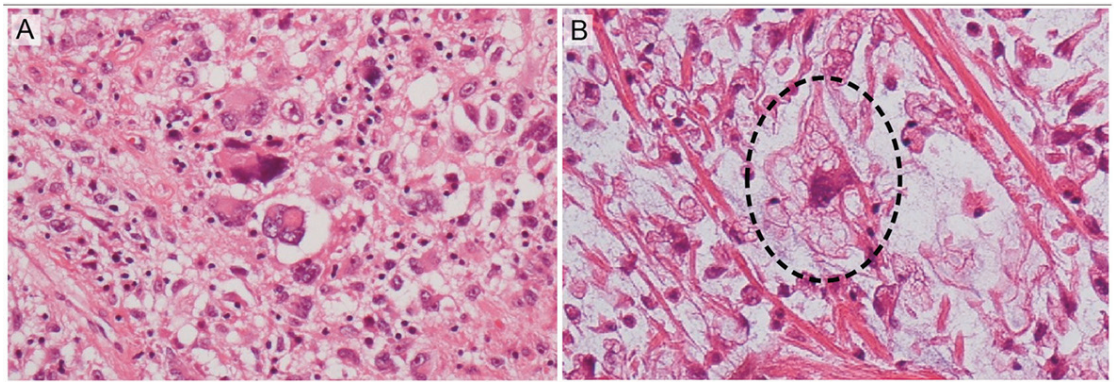

Figure 4 **Histopathological findings.**
A: Multinucleated giant cells, showing various pleomorphisms, are evident (hematoxylin and eosin staining [H&E], ×200 magnification).
B: A Lipoblasts can be seen (H&E ×400). The case was diagnosed as a pleomorphic liposarcoma originating from the preperitoneal fat.

margins were negative (Figure 4).

The patient's postoperative course was uneventful, and he was discharged on the fifth postoperative day. Following surgery, follow-up blood samples were taken and CT examinations performed every 6 months. The patient remains alive five years after the operation without disease recurrence.

Discussion

The growth pattern of a liposarcoma is largely expansive and there is little tendency of the tumor to infiltrate surrounding tissues. Indeed, patients report few subjective symptoms until the surrounding organs are compressed. These tumors are therefore often very large by the time they are detected. The well-differentiated type, according to the WHO histological classification, accounts for 46% of diagnosed liposarcomas, with the frequencies of the myxoid, round cell, pleomorphic, and dedifferentiated types being 18%, 18%, 10%, and 8%, respectively; thus, the pleomorphic type is relatively rare.[5] To date, pleomorphic liposarcomas have been reported in the limbs, retroperitoneum, abdominal wall, chest wall, mesentery, pelvic cavity, spermatic cord, fallopian tube, mediastinum, parietal pleura, pericardium, spinal cord, and head and neck.[6-11] Although the preperitoneal fat origin of the liposarcoma described in the present case cannot be absolutely confirmed by our examination, its location makes it an extremely rare case. As far as we aware, there were four reported cases of liposarcoma of abdominal wall origin, but none of them were derived from preperitoneal fat, and the histology was myxoid in two cases[12,13], well differentiated in two cases [14,15], with no pleomorphic type. Although it cannot be completely ruled out, it would be that the retroperitoneal origin is frequently encountered due to the presence of various adipose

tissues associated with retroperitoneal organs (perirenal fat, pre-pancreatic fat, etc.). In contrast, preperitoneal fat origin is very rare because of a homogeneous fat layer located between the muscular layer and the peritoneum, lacking such diversity.

The prognoses for patients with the well-differentiated and myxoid types of liposarcomas are quite favorable, with 5-year survival rates of 93% and 92%, respectively. In contrast, the round cell, pleomorphic, and dedifferentiated types are highly malignant, with reported 5-year survival rates of 74%, 59%, and 44%, respectively.[5] Hornick et al. examined 57 cases of pleomorphic liposarcoma and identified the following factors (commonly linked with pleomorphic liposarcomas) to be associated with poor outcomes: occurrence in the center of the body (not the extremities), occurrence below the fascia, diameter larger than 10 cm, presence of many mitotic cells, presence of necrotic features, and pleomorphism of the epithelium.[12]

Ultrasound examination, CT, and MRI are used to diagnose liposarcomas. The tumors frequently appear as heterogeneous, hyperechoic masses on ultrasonography. CT shows the tumor to have a density similar to that of fat; the presence of inhomogeneous and diverse mass interiors increases the likelihood of liposarcoma. The higher the degree of differentiation, the lower the CT signal intensity, and the poorer the degree of differentiation, the higher the CT intensity. MRIs of well-differentiated liposarcomas often show high signal intensities in T1- and T2-weighted images, and poorly differentiated liposarcomas often show low signal intensities in T1-weighted images and high signal intensities in T2-weighted images. Brenner et al. reported a mean maximum SUV of 3.6 for 54 liposarcomas examined using PET-CT.[13] Maximal SUVs tended to be higher in the following order of liposarcoma types: well-differentiated, followed by myxoid, dedifferentiated, and pleomorphic; the maximum SUV tended to be proportional to the malignancy of the tumor. Consistent with Brenner et al. report, our case showed a strong accumulation of fluorodeoxyglucose. According to the same report, the recurrence-free survival was reported to be significantly shorter in patients with maximum SUV values ≥ 3.6, compared with cases with maximum SUV values < 3.6. The absolute maximum SUV values may be useful as prognostic predictors.[13]

The histopathological examination of our patient's tumor revealed a marked pleomorphism of spindle-shaped lipoblasts in hematoxylin and eosin-stained specimens. In addition to round and spindle-shaped cells of varying sizes, presence of multinucleated giant cells with deeply stained nuclei was observed. The differential diagnosis for pleomorphic liposarcomas may include pleomorphic leiomyosarcoma and pleomorphic malignant schwannoma. However, the presence of pleomorphic lipoblasts is characteristic of a pleomorphic liposarcoma. Immunohistochemical studies involving assessments for the presence of markers such as S-100, cytokeratins, CD34, and desmin can be performed to rule out other diseases.

Surgical excision is the first choice of treatment. Liposarcomas often present as tumor cells covered with a flattened pseudocapsule. These tumors require complete resection, without exposing the capsule. A prognostic analysis of liposarcoma margin characteristics reported 12-year disease-specific survival rates of 74% associated with negative margins, 68% for cases with positive microscopic margins, and 25% for those with positive macroscopic margins.[5] Although our case involved a pleomorphic liposarcoma and had a poor prognosis, the patient achieved 5 years of recurrence-free survival due to the complete resection of his tumor.

Conclusions

In conclusion, we reported a case of pleomorphic liposarcoma originating from the preperitoneal fat. Although pleomorphic liposarcomas have poor prognoses, an improved long-term prognosis may be expected following the complete resection of the tumor, as in the present case. For tumors that may originate from the preperitoneal fat, a surgical excision is required.

Abbreviations: WHO, World Health Organization; CT, computed tomography; SUV, standardized uptake value; MRI, magnetic resonance imaging; PET, positron emission tomography.

Authors' contributions: Yasumasa Yoshioka wrote the manuscript; Tadao Tokoro designed the study; Yusuke Makutani, Ogawa Ryotaro and Hokuto Ushijima analyzed the data; Masayoshi Iwamoto, Toshiaki Wada, Koji Daito studied the relevant literature and reviewed the data; Kazuki Ueda and Junichiro Kawamura edited the manuscript and figures. All authors read and approved the final manuscript.

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