

Case Report

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Primary pleomorphic liposarcoma of fallopian tube with recurrence: a case report and review of the literature

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Abstract: Background. Liposarcoma, which develops in adipose tissue, is one of the most common soft tissue sarcomas. It appears mostly in the lower limbs, particularly in the thigh and limb girdles, followed by the upper extremities, thoracoabdominal wall, and the internal trunk and retroperitoneum. Pleomorphic liposarcoma (PLS), a rare subtype of liposarcoma is considered a high-grade malignancy.

Case presentation. We present a case of primary PLS in the left fallopian tube of a 47-year-old female. Ten months previously, she was diagnosed with PLS of the left fallopian tube in another hospital and had a wide excision of left fallopian tube mass, including total abdominal hysterectomy and bilateral salpingo-oophorectomy for left fallopian tube PLS. Presently, she has developed a recurrence and metastasis of PLS in the pelvic and abdominal cavities.

Conclusion. This may be the first case of primary fallopian tube PLS, wherein the prognosis of this patient was poor due to the high-grade malignancy of PLS.

Keywords: Pleomorphic liposarcoma; Liposarcoma; Surgery

1 Introduction

Liposarcoma, which develops in adipose tissue, is one of the most common soft tissue sarcomas. Pleomorphic liposarcoma (PLS), a rare subtype of liposarcoma, is considered a high-grade malignancy. The study of Gebhard et al. [1], which involved 63 PLS cases, suggested that it is usually located in the proximal extremities, such as the lower extremities (36.5%), and particularly in the thigh (28.5%) and limb girdles (17.5%), the upper extremity (16%), and thoracoabdominal wall (9.5%), and internal trunk (20.5%). Some uncommon sites including the mediastinum [2], parietal pleura [1], pericardium [3], and head and neck region [4] were also found. Herein, we report what is possibly the first case of PLS in the left fallopian tube in a 47-year-old woman.

2 Case report

A 47-year-old woman visited the hospital with a 2-month history of dull pain in the left lower quadrants of the abdomen. Ten months previously, she first visited a doctor at another hospital because of the discontinuous, dull pain in left lower abdomen, but without vaginal bleeding and fluids. At that time, there was a newborn head-sized mass in the hypogastrium. The transabdominal ultrasound showed an 11.3 x 8.7 x 10.8 cm-sized mixed echogenic mass with irregular morphology in the front of the uterine body; there were a small number of blood flow signals. She had undergone wide excision of the left fallopian tube mass, including total abdominal hysterectomy and bilateral salpingo-oophorectomy for the left fallopian tube mass. She was diagnosed with PLS of the left fallopian tube at another hospital through histopathology of the surgical tissues. She then received a 21-day chemotherapy regimen with ifosfamide (2 g, days 1–3; intravenous injection (ivgtt)) and epirubicin (60 mg, day 1; 50 mg, day 2; ivgtt); the chemotherapy progressed very well, but

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she then refused subsequent treatment for the economic and familial reasons. She did not present any history of alcohol consumption, smoking, transfusions, other surgeries, various communicable diseases, or chronic diseases. Her menstrual cycle was regular, with a mean of 30 days, moderate flow, a dark red color, and no history of dysmenorrhea. Although she had three pregnancies, she had given birth to only one child.

The relevant results of her first surgery at another hospital were as follows: Gross findings of the specimen revealed a smooth and encapsulated mass, measuring 10 x 10 x 9 cm at initial presentation, without adhesion to the surrounding tissues. Histopathological analysis (Figure 1) revealed multiple smooth muscle cells, lipoblasts, and pleomorphic cells; the smooth muscle cell area had been invaded by pleomorphic cells, especially spindle cells. The lipoblasts were characterized by heterotypic, hyperchromatic nuclei with uni/multivacuolated cytoplasm. There were various types of pleomorphic cells, including mono/multinucleate giant cells, spindle cells, and bizarre cells. Immunohistochemistry analysis showed that CD34, CD117, CD10, DOG, and S-100 protein were negative, the index of Ki-67 was 20%–30%. There was no evidence that suggested tumor necrosis and metastasis. As her first surgery had been performed at another hospital, we were mainly able to provide textual descriptions as supportive evidence, in the absence of some figures.

Various results of the time in our hospital were follows.

Physical examination showed a soft, flexible mass apparent in the left lower quadrant of the abdomen. Laboratory data upon admission were not unusual. Levels

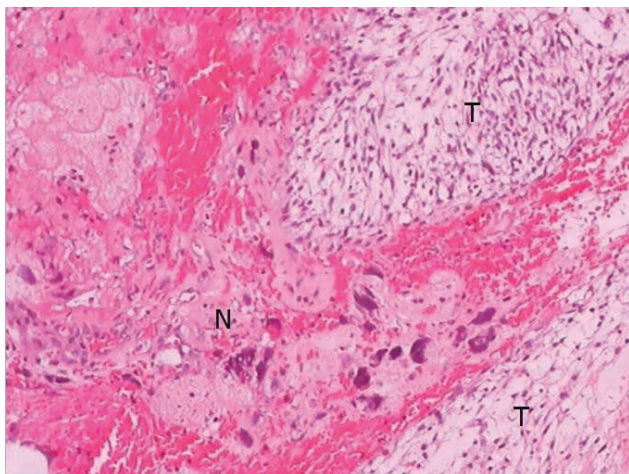


Figure 1: Histopathological slide of the first surgery of this patient. The oviduct smooth muscle cell area had been invaded by pleomorphic cells, in particular, spindle cells. N indicates necrosis of the oviduct smooth muscle area; T: tumor area, which contained multiple spindle cells.

of tumor markers, including carcinoembryonic antigen (CEA), carbohydrate antigen (CA) 19-9, and CA-125 were all within normal limits. There was no apparent abnormality observed in echocardiography. Sonography (transabdominal ultrasound) (Figure 2A) showed an 11.3 x 7.9 x 11.3 cm-sized mixed echogenic mass in the left lower quadrants of the abdomen, the boundary of which was relatively clear, and there was a small number of blood flow signals. A preoperative computed tomography (CT) (Figure 2B) scans upon admission showed multiple small lymph nodes in the retroperitoneum, and multiple lesions in the left side of pelvic cavity and the left lower abdominal mesenteric area. They were considered metastatic tumors. Her thorax CT scans (Figure 2C) showed a funicular image in the inferior lobe of the left lung.

Twenty-one days of chemotherapy with ifosfamide (2 g, days 1–4; ivgtt) and epirubicin (60 mg day 1; 50 mg day 2; ivgtt) was administrated initially; she then underwent a liposarcoma cytoreduction with retroperitoneal neoplasm resection, colon sigmoideum and partial small bowel resection, appendectomy and enterolysis for the recurrence of left oviducal PLS with abdominal and pelvic cavity metastasis.

The pathological results of the second surgery were as follows.

Gross findings of the specimen revealed that there was a 4.0 x 3.0 x 3.0 cm-sized smooth and round mass with a gray cut surface at the left side of the pelvic wall, and a 5.0 x 4.0 x 3.5 cm-sized mass at the sigmoid mesocolon with the same characteristics as the left mass, and multiple protruded lesions with smooth surfaces and a maximum diameter of 0.3 cm at the right side of the pelvic wall and the appendiceal wall of the iliac fossa. Reports on histopathology (Figure 3) of these masses revealed reports similar to previous reports. She was diagnosed with recurrent PLS with metastasis.

At present, the patient has been discharged from the hospital, and has refused further treatment with either radiotherapy or chemotherapy. Based on the present follow-up information, she has survived with metastatic tumors and without any ongoing treatment.

Ethical approval: The research related to human use has been complied with all the relevant national regulations, institutional policies and in accordance the tenets of the Helsinki Declaration, and has been approved by the authors' institutional review board or equivalent committee.

Informed consent: Informed consent has been obtained from all individuals included in this study.

3 Discussion

Liposarcoma is very rare in the female reproductive system, especially in the fallopian tubes. Oishi et al. reported the first case of liposarcoma (histopathologically diagnosed as well-differentiated liposarcoma) arising

from the fallopian tubes, but the primary site was the uterus, not the fallopian tubes [5]. PLS is a type of pleomorphic, highly malignant liposarcoma with various unique lipoblasts observed in histopathology tests. It is rare and only accounts for 5% of liposarcoma cases [1]. It has been reported that PLS originates primarily in the orbit [6] and uterus [7]. The present report may present the first case of primary fallopian tube PLS. PLS is considered to harbor the highest possible grade of malignancy, with the poorest prognosis among the liposarcomas [1]. In addition, it has been reported [8, 9] that the 1-, 3-, and 5-year survival rates of PLS are 93%, 75%, and 29%, respectively. Therefore, it is extremely important that PLS be diagnosed early and treated appropriately. However, because it is characterized as the growth of a progressively painless mass, it is easily overlooked until the mass is sufficiently large or other manifestations are observed. In the present case, the patient first visited a physician because of a discontinuous, dull pain in left lower abdomen. At that time, there was a newborn head-sized mass in the hypogastrium. However, specific and effective diagnosis methods do not yet exist; its diagnosis still relies on pathological analysis of the surgical mass. At present, there are no globally standardized treatment approaches [10]. Radical surgical resection is the best and main treatment for PLS; chemotherapy and radiotherapy as a multi-modality treatment strategy still remain controversial. This patient experienced recurrence and metastasis ten months after radical surgical resection of PLS in the left fallopian tube.

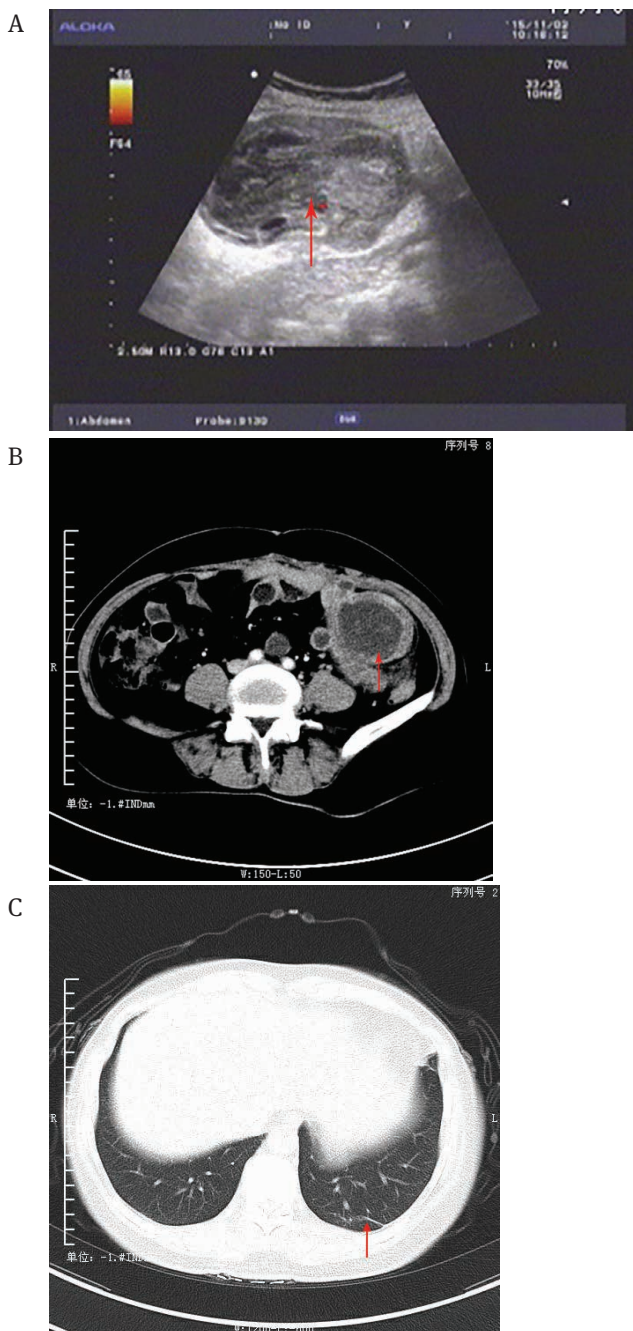


Figure 2: Auxiliary examinations of the patient, the abnormal sites have been labeled by red arrows. There was a mixed echogenic mass in her left lower quadrant of the abdomen on sonography (2A, on the left). There was a large lesion in the left lower abdominal mesenteric area as observed on abdominal CT images (2B, middle). Her thorax CT scans showed a funicular image in the inferior lobe of the left lung (2C, right).

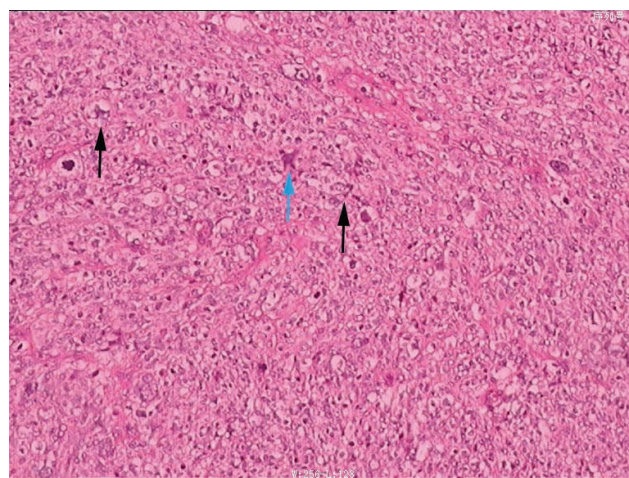


Figure 3: Histopathological slide of the pelvic wall mass of the second surgery. It revealed the lipoblasts and pleomorphic cells. The black arrows indicate lipoblasts, which were characterized by heterotypic, hyperchromatic nuclei with uni/multivacuolated cytoplasm. The blue arrow indicates one of the pleomorphic cells, polymorphonuclear cell.

A study by Hornick *et al.* [9] involving 57 PLS cases reported that a shorter distance between the mass and the center of the trunk correlated with poorer prognosis. Larger mass size (more than 10 cm) correlated with a deeper location, necrosis of the tumor, poorer prognosis, likelihood of relapse, and decreased survival time. This case nearly met those criteria both upon primary and recurrent LPS detection. The prognosis for this patient is not optimistic, even though she remains alive with metastatic tumors three months after discharge.

4 Conclusion

PLS often presents with relapse and metastasis, and the survival rate is low. We report the case of a 47-year-old woman with primary PLS recurrence of the left fallopian tube who initially underwent radical surgical resection of the tumor, and subsequently underwent a cytoreductive surgery for the recurrence and metastasis of PLS 10 months after the initial surgery. However, the prognosis for this patient was not optimistic. Increasing the survival rate of PLS patients and even curing PLS remain challenges for every medical professional.

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