A case of retroperitoneal liposarcoma after delivery with expression of estrogen receptor: Report of a case

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INTRODUCTION: Liposarcoma is one of the most common soft tissue sarcomas; however, early diagnosis is rare as the tumor remains difficult and unpalpable for a prolonged period of time.

PRESENTATION OF CASE: Here we report the first case of retroperitoneal liposarcoma associated with pregnancy and estrogen receptor expression. A 34-year-old woman experienced persistent abdominal distension after her first delivery. Computed tomography (CT) and magnetic resonance imaging (MRI) revealed a large (40 cm × 35 cm), solid, palpable abdominal mass with fat attenuation displacing the ascending colon and the right kidney to the left. Laparotomy and an en-bloc resection of the tumor were performed; further, right nephrectomy and adrenalectomy were required. Histopathology showed a well-differentiated liposarcoma; approximately 10–20% of the tumor cells were ER-positive.

DISCUSSION: Retroperitoneal liposarcoma associated with pregnancy is an extremely rare occurrence. Surgical resection is unquestionably the first choice of treatment, but complete resection is sometimes impossible due to the volume and depth of invasion of the tumor. In such cases, additional therapy for liposarcoma is important to improve prognosis. Thus, this report highlights the need for further research into hormone therapy.

CONCLUSION: Retroperitoneal liposarcoma has a high local recurrence rate due to the difficulty in complete surgical resection; therefore, additional hormone therapy is important for improving the prognosis.

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The retroperitoneal mass weighed 7200 g and measured 43 cm × 40 cm × 13 cm (Fig. 2). Histopathological examination of the surgical specimen showed a well-differentiated liposarcoma (Fig. 3). Immunohistochemistry revealed approximately 10–20% of the tumor cells to be estrogen receptor (ER)-positive, with negative results for the progesterone receptor (PR) (Fig. 4). The patient recovered uneventfully and was discharged from the hospital on the 15th postoperative day. She and her husband were planning to try for another baby, so postoperative adjuvant therapy including hormone therapy was not undertaken. No recurrence was detected at the 3-year follow-up.

3. Discussion

Retroperitoneal sarcomas represent approximately 10–15% of all soft tissue sarcomas, which themselves are rare, accounting for only 1% of all malignancy [3]. The peak incidence of liposarcoma occurs from 40 to 60 years of age. The tumor is typically aggressive, with reported 5-year survival rates ranging from only 23% to 46% [4]. Lewis et al. [5] reported high local recurrence rates for retroperitoneal sarcoma, ranging from 40% to 80%. As the retroperitoneal space is easily distended by the enlarging tumor, the neoplasia remains inactive and unpalpable for prolonged period of time. A retroperitoneal mass may produce a wide range of signs and symptoms due to the compression and infiltration of the surrounding organs: localized pain associated with neurologic symptoms due to neuroplexus involvement, vomiting due to gastric compression, constipation, hydronephrosis, uremia due to renal disorder, and ureteric compression. Izumi et al. [6] reported that the most common symptom of retroperitoneal sarcomas is abdominal pain (25.8%), followed by abdominal distension and palpable mass, while 22.1% of the cases develop without symptoms. In the present case, the first symptoms were continuous abdominal distension and respiratory discomfort lasting throughout the pregnancy without improvement, suggesting that the mass existed and grew during the pregnancy. Ultrasonographic examinations performed as routine check-ups during the perinatal period did not detect the retroperitoneal tumor in this case. Hence, it is recommended that a transabdominal ultrasonographic examination (or MRI if necessary) be performed in cases where a pregnant woman complains of abdominal distention or respiratory discomfort.

We reviewed the literature on liposarcoma associated with pregnancy and found 16 cases including ours. Their clinical courses are summarized in Table 1 [1,3,7,8,10–18]. The mean age was 31 years (range, 15–44 years). Four cases of liposarcoma were discovered during the first trimester, 10 during the second or third trimester, and 2 within the 1 year postnatal period. Only 2 patients diagnosed in the first trimester were surgically treated during pregnancy; in all other cases, surgery was performed after delivery.

Liposarcoma is classified into five histological subtypes: well-differentiated, round-cell, myxoid, pleomorphic, and de-differentiated (WHO classification, 2002). There were 8 cases of myxoid type and 4 cases of well-differentiated type, both of which were classified as low-grade. Twelve cases were large tumors exceeding 15 cm in diameter. According to the reports, surgery was the only treatment given during pregnancy; however, radiotherapy to the lower extremity after delivery [7] and chemotherapy in combination of mesna, doxorubicin and ifosfamid after surgical excision [8] have been reported. In terms of prognosis, out of the 4 patients diagnosed in the first trimester, 2 survived following surgery under general anesthesia at 13th week of pregnancy. However, all patients for whom surgery was performed after delivery died. Therefore, surgical resection should be considered as soon as possible, even if the initial diagnosis is made during pregnancy.

The effect of pregnancy on tumors is a major concern. In our patient, a relationship existed between tumor growth and pregnancy, and immunostaining was ER-positive. Cantin and
### Table 1
Clinicopathological characteristics of pregnancy-associated liposarcoma.

| Author          | Age | complaint          | Gestation at diagnosis (W) | Location    | Surgery                          | Tumor subtype | Size (Weight) | Prognosis                      |
|-----------------|-----|--------------------|-----------------------------|-------------|----------------------------------|---------------|---------------|--------------------------------|
| Pierandrea D.J. et al. [1] | 41  | Weight loss        | 34                          | Retroperitoneum | 36 weeks of gestation concurrently with CS | Myxoid        | 23 cm         | Died after 8 months           |
| Jeng C-J. et al. [3]       | 33  | Unknown            | 12                          | Retroperitoneum | 36 weeks of gestation concurrently with CS | Myxoid        | 25 × 20 cm     | Recurrence after 4 months postop. |
| Matsuda S. et al. [7]      | 28  | Pain in left thigh | 29                          | Left thigh    | 5 weeks after CS at 33 weeks of gestation | Myxoid        | 15 × 10 × 5 cm | Free of disease 5 years postop.  |
| Tebes S. et al. [8]        | 22  | Abdominal pain     | 13                          | Retroperitoneum | 3 weeks after VD at 29 weeks of gestation | Pleomorphic   | 20 cm         | Died after 2 months           |
| Foruhan B. [10]           | 28  | None               | 28                          | Retroperitoneum | 38 weeks of gestation concurrently with CS | Myxoid        | 30 cm (2.678 kg) | Free of disease 18 months postop. |
| Yamamoto T. et al. [11]    | 29  | Palpable mass      | 29                          | Right thigh   | none (chemoradiotherapy after CS) | Myxoid        | 8 × 8 × 5 cm    | Remission 13 months postop.     |
| Kurogouchi A. et al. [12]  | 30  | Palpable mass      | 12                          | Retroperitoneum | 13 weeks of gestation (CS at 37 weeks of gestation) | Myxoid        | 34 × 14 × 14 cm | Alive 4 years lataer with operation for recurrence Free of disease 1 year postop. |
| Lopes R.I. et al. [13]     | 33  | None               | 13                          | Retroperitoneum | 13 weeks of gestation (CS at 37 weeks of gestation) | Well-differentiated | 22 × 20 × 20 cm(3.75 kg) | Free of disease 1 year postop.     |
| Jafari K. et al. [14]      | 15  | Chest pain         | 31–32                       | Mediastinum   | none (chemoradiotherapy after VD) | Anaplastic    | unknown        | Remission                      |
| Carrol F. et al. [15]      | 23  | Respiratory distress | 32–33                      | Left pleura   | 32–33 weeks of gestation concurrently with CS | Mixed-type    | 29 × 15 × 15 + 21 × 12 × 8cm(4.46 kg) | Unknown                        |
| D-Garcia O.F. et al. [16]  | 35  | Palpable mass      | 36                          | Retroperitoneum | after delivery (unspecified) | Well-differentiated | 52 × 40 × 35 cm (12.5 kg) | Free of disease 1 year postop. Died after 1 month |
| Rouskova L. et al. [17]    | 32  | Weight loss        | 34                          | Retroperitoneum | 3 days after delivery | Pleomorphic  | unknown        |                               |
| Masuda T. et al. [18]      | 34  | Unknown            | Diagnosis at delivery       | Retroperitoneum | 3 months after delivery | Myxoid        | 7.5 × 7.0 × 7.1 cm | Free of disease 8 years postop. Free of disease 3 years postop. |
| Our case                  | 34  | Abdominal distension | 32 weeks after delivery    | Retroperitoneum | 9 months after delivery | Well-differentiated | 43 × 40 × 13cm(7.2 kg) |                                 |

CS, cesarean section; ID, induced delivery; VD, vaginal delivery.
McNeer [2] determined that pregnancy does not adversely affect the prognosis of the tumor. However, they suggested that an estrogen–progesterone environment possibly has a favorable influence on the natural history of sarcoma, and the value of additive hormone therapy in the management of metastatic sarcoma should be explored. Matsuda et al. [7] reported that soft tissue sarcoma can grow rapidly during pregnancy. Xiao-Qiu Li et al. [9] who analyzed 120 soft tissue sarcomas including 31 liposarcomas for ER expression reported that neither gender nor age have a significant influence on the ER expression status, but further studies to assess the hormonal dependency of ER-positive mesenchymal neoplasms and potential therapeutic applications would be of clinicopathological significance. Retroperitoneal liposarcoma associated with pregnancy is an extremely rare occurrence, and no case investigating hormone receptors has been reported in the literature. Surgical resection is unquestionably the first choice of treatment for retroperitoneal liposarcoma, but a complete resection can sometimes be impossible due to the volume and depth of invasion of tumor. Therefore, additional therapy for liposarcoma is important to improve prognosis; thus, this report highlights the need for further research into hormone therapy.

**Conflicts of interest**

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**Ethical approval**

This manuscript has been met ethical approval.

**Consent**

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

**Author contribution**

Hiroaki Kasashima: writing the paper, doctor in charge. Yoshio Yamasaki: writing the paper, doctor in charge, guarantor. Yoshikazu Morimoto: data interpretation. Yusuke Akamaru: data interpretation. Keigo Yasumasa: doctor with this patient’s surgery. Tsutomu Kasugai: pathological analysis. Yasuyuki Yoshida: pathological analysis.

**Guarantor**

The guarantor of this manuscript is Yoshio Yamasaki.

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