Fulminant course of metastatic liposarcoma after delivery - case report

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Summary

Abdominal liposarcoma is a rare tumor of uncertain prognosis. Radical surgery is possible in about two-thirds of the patients, and the prognosis of patients with inoperable tumors is dismal. Only a few cases of liposarcoma complicating pregnancy have been documented. We report a case of a patient who was diagnosed with metastatic abdominal liposarcoma during the third trimester of the pregnancy. After induced vaginal delivery, palliative surgery was performed and one cycle of systemic combination chemotherapy was administered. Despite the multimodality treatment the patient died of progressive disease within one month after diagnosis. Autopsy revealed high-grade pleomorphic liposarcoma arising from the retroperitoneum with liver and lung metastases.

Key words: Liposarcoma; Pregnancy; Retroperitoneum.

Introduction

Liposarcomas are rare tumors that comprise about 15% of all cases of soft tissue sarcoma and 50% of retroperitoneal sarcomas [1]. Radical surgery, the only curative therapy, is possible in 60-70% of patients, but the tumor eventually recurs in half of these cases [1]. Only about a dozen cases of liposarcoma complicating pregnancy have been documented in the literature [2].

Case Description

A 32-year-old woman, gravida 4, para 3, was admitted at 34 weeks gestation because of weight loss, vomiting and cough. The previous three pregnancies had been uneventful and her personal history was not contributory. The course of the present pregnancy until week 32 was uncomplicated, with the patient complaining only about mild discomfort in the hypogastrium.

On physical examination, anemic coloration and tachycardia were noted. The uterus was normotonic, and the cervix was closed. Ultrasonographic examination revealed a normal fetus in vertex presentation with biometrical parameters corresponding to the gestational age. Laboratory findings showed moderate anemia, thrombocytosis, hypoproteinemia, and high C-reactive protein. A mass in the left lung was detected on chest X-ray. Multiple echogenic lesions in the liver and a large cystic and solid formation were identified by abdominal ultrasound. Because of the suspected diagnosis of metastatic malignancy, labor was induced at 35 weeks of gestation, resulting in an uncomplicated vaginal delivery of a healthy female infant (2,300 g). Abdominal computed tomography scan performed after delivery revealed a large tumor in the left mesogastrium and hypogastrium, and muliple liver metastases (Figure 1).

On the third day after delivery the patient presented with signs of intestinal obstruction. Operative revision was indicated, and during surgery an inoperable large cystic tumor mass infiltrating the left retroperitoneum and mesentery was found. Only palliative debulking of the tumor mass consisting of removal of necrotic contents of the cystic mass was possible.

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Preliminary results of the histological examination suggested poorly differentiated sarcoma. Systemic combination chemotherapy with doxorubicin (140 mg in 24-hour infusion) and ifosfamide (3 g in 24-hour infusion for 5 days) with mesna was administered starting on the 12th postoperative day. The condition of the patient did not improve, and nine days later she was diagnosed with pneumonia and neutropenia. Despite intensive antibiotic treatment the condition of the patient deteriorated, and she died of respiratory failure on the 26th day after surgery.

At autopsy, lung edema, multiple liver metastases, a solitary metastasis in the lower lobe of the left lung (9 cm) and tumor infiltrating the peripancreatic tissue and left kidney were found. Histologic examination revealed high-grade pleomorphic liposarcoma.

Discussion

Among six patients with abdominal liposarcoma complicating pregnancy reported in the literature, three patients died within one year of diagnosis and local recurrence was diagnosed in another patient (Table 1). In contrast, the outcome in patients with pregnancy complicated by liposarcoma arising in the extremities has been largely favorable [2]. Retroperitoneal sarcomas are distinguished from intraabdominal liposarcomas arising in the mesentery [3], but in patients with advanced voluminous tumors, as in the present case, this distinction may be difficult, if not impossible. Unlike liposarcoma located in the extremities, retroperitoneal or abdominal primary may complicate vaginal delivery. Cesarean section has been used, but vaginal delivery was possible in selected cases.

The fulminant course of liposarcoma with synchronous metastases to the lung and liver in the present patient is rather unusual. Pregnancy might have contributed to the unusually rapid progressive course. Surgery resulted only in short-term palliation, and survival was even shorter than in both cases of fatal retroperitoneal sarcoma complicating pregnancy reported so far [4, 5]. In a large series

Reference	Patient age (years)	Location of the primary tumor	Histology	Gestation age at diagnosis	Delivery	Therapy	Patient outcome
(5)	41	retroperitoneal	myxoid	34 weeks	cesarean	resection	died
(6)	31	retroperitoneal	(high-grade) myxoid	third trimester	(36 weeks) cesarean	resection	(8 months) local recurrence after 12 months,
(3) (4)	32 22	mesentery retroperitoneal	myxoid not specified	7 months 26 weeks	vaginal vaginal (29 weeks)	resection resection + chemotherapy	repeat resection alive (7 years) died (4 months)
(7)	28	retroperitoneal	myxoid	39 weeks	cesarean	(doxorubicin + ifosfamide) resection	alive (18 months)
present report	32	mesentery	pleomorphic (high-grade)	34 weeks	(39 weeks) vaginal (35 weeks)	palliative resection + chemotherapy (doxorubicin + ifosfamide)	died (1 month)

Table 1. — Summary of reports of abdominal liposarcoma complicating pregnancy.

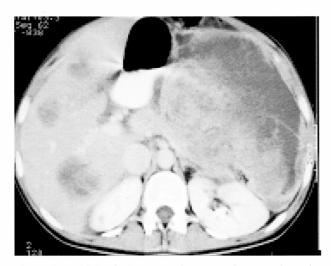


Figure 1. — Abdominal computed tomography scan showed a voluminous cystic and solid tumor in the left mesogastrium and hypogastrium and multiple hepatic metastases involving both lobes.

of patients with retroperitoneal sarcoma reported by Jaques *et al.* [1], only three of 114 patients had synchronous metastases and none of 57 patients with liposarcoma developed liver metastases. The prognosis of retroperitoneal liposarcoma in pregnancy seems to be inferior compared to other cases of this tumor.

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