

A Rare Case of Leiomyosarcoma with Hemoperitoneum in Pregnancy

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Abstract

Uterine leiomyosarcoma during pregnancy is an extremely rare event. We report a retrospectively diagnosed case of a ruptured primary uterine leiomyosarcoma with hemoperitoneum diagnosed during pregnancy. The patient came to the emergency room (unbooked with us, taking treatment at the other hospital) with G4P1A2L0 with GDM + pregnancy-induced hypertension (PIH) and ultrasonography (USG) diagnosed large fibroid with degeneration (19 cm × 13.2 cm × 13.2 cm) at 30.5 weeks of gestation, breech presentation with complaints of pain in abdomen and vomiting. An initial conservative treatment was started considering prematurity with fibroid degeneration. Due to severe pain in abdomen and history of PIH, USG was repeated after 2 days of admission which showed hemoperitoneum, so the decision for emergency preterm LSCS was taken. During cesarean section, a ruptured sessile uterine fibroid was removed and hemoperitoneum was drained. Histopathological diagnosis of the tumor revealed a poorly differentiated uterine leiomyosarcoma. The patient underwent laparotomy after weeks with radical hysterectomy and received adjuvant chemotherapy. The diagnosis and suspicion for leiomyosarcoma during pregnancy at young age stand difficult, and the treatment options seem to be reduced in pregnant women and are mainly dependent on the patient's condition as well as the gestational age at the time of presentation.

Key words: Degenerated fibroid, Hemoperitoneum, Leiomyosarcoma, Pregnancy, Radical hysterectomy

INTRODUCTION

Uterine leiomyosarcomas (LMS) are rare, highly malignant neoplasms that make up about 1% of all uterine malignancies.^[1] The risk of local recurrence and metastasis is high, with reported 5-year survival rates ranging between 12% and 25%. The median age for women with LMS is after childbearing age (between 43 and 53 years). Total abdominal hysterectomy and bilateral salpingo-oophorectomy are considered the standard therapy for LMS of the uterus.^[2,3] The role of conservative fertility-sparing surgery in young women remains controversial.^[4] Uterine LMS associated with pregnancy is even rarer and only a few cases are reported in the literature.^[4-6] Here, we report a case of a patient with retrospectively diagnosed uterine LMS during the third trimester of pregnancy.

CASE REPORT

The patient came to the ER with G4P1A2L0 with GDM and ultrasonography (USG) diagnosed large degenerated fibroid at 30.5 weeks of gestation with breech presentation with pain in abdomen and vomiting associated with pregnancy-induced hypertension (PIH). The patient came to us for the first visit, previously taking treatment at other hospitals. On taking detailed history, the patient was G4P1A2L0 at 30.5 weeks:

LMP: 16/12/16, EDD: 23/9/17.

Obstetric history was as follows:

- G1: Missed abortion at 10 weeks followed by D and C 10 years back.
- G2: Preterm vaginal delivery at 6 months, 7 years back and the baby died after 4 days.
- G3: Missed abortion at 10 weeks f/b D and C, 6 years back.
- G4: Present pregnancy, spontaneous conception.

Medical history: The patient was diagnosed with GDM in the second trimester on tablet metformin 500 mg thrice daily

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(controlled blood sugar levels). Diagnosed with PIH 2 days back was on tablet labetalol 200 mg thrice daily for 2 days. The patient was given antenatal steroid coverage 2 days back.

There was no significant family history.

On admission, the patient was conscious and oriented, afebrile, pulse rate was 84/min, blood pressure (BP) 160/80 mmHg; on P/A examination, uterus size was 34 weeks, relaxed, fibroid was palpable separately and slightly tender. Gentle P/S showed os closed, no discharge.

Initially, conservative management was planned in view of PIH + GDM and prematurity. The patient was given injectable labetalol for uncontrolled PIH, injection PCM, and injection Rantac/Emset. USG was done on admission which showed: Single live intrauterine pregnancy of 31.2 weeks maturity, breech presentation, EBW: Approximately 1.79 kg, anterior placenta, normal Doppler findings, and a large subserosal/intramural fibroid of 19 cm × 13.2 cm × 13.2 cm. Laboratory investigation showed - serum glutamic oxaloacetic transaminase: 18, lactate dehydrogenase: 196, serum glutamate-pyruvate transaminase: 15, CREAT: 0.4, uric acid: 6.1, HB: 10.1 g%, and blood group: O positive. Blood sugar charting was done and was managed along with the physician; insulin was started.

Pain and vomiting were controlled for 1 day, but dull aching pain was still persistent. After 2 days of admission, the pain aggravated and not relieving with IV painkillers and repeat PIH profile showed deranged coagulation profile, altered liver function tests, raised urine ketone bodies, and unstable blood sugar and BP. USG was done which showed live intrauterine fetus with hemoperitoneum. The patient was reviewed by physician and anesthetist and was taken for emergency LSCS under general anesthesia. She delivered male baby weighing 1.6 kg breech footling presentation with meconium stained liquor. Baby admitted in NICU for 3 weeks and discharged in healthy condition.

The intraoperative findings are as follows:

1. After opening abdomen in layers, parietal peritoneum was visualized and blood shown in the peritoneal cavity. Dark red blood presents in the cavity on opening the peritoneum suggestive of hemoperitoneum. Approximately: 2500 mL of blood was suctioned out.
2. Baby was delivered and uterus sutured. Uterus was bulky and exteriorized. Small 3 cm × 2 cm sessile subserosal fibroid shown on posterolateral side of uterus. A tear was visualized just below the right tuboovarian ligament and extending laterally toward the broad ligament. Large uterine fibroid with degenerative material and clots was coming out. Around 1 L/1 kg of fleshy pieces removed.

3. All the degenerative tissue manually removed by finger manipulation into the tear which was going deep posteriorly to the uterus. Clots of dark blood and degenerative fibroid found lying in the peritoneum, which were removed manually. Degenerated tissue removed to a great extent.
4. Right tube and ovary were very edematous. Tear in the right mesosalpinx and near ampulla of the right fallopian tube shown which was sutured. The left tube and ovary, omentum appeared normal. Hemostasis was achieved and specimen was sent for HPE. The patient was transfused 3 units PRBC and 2 units FFP and was closely monitored in ICU for 24 h with iv fluids, higher antibiotics and hemostatics with DVT pump.

The only positive finding in the USG performed on day 4 post operatively was borderline hepatosplenomegaly. Postpartum bulky uterus with subserosal fibroid with 8.4 cm × 8.4 cm × 8.4 cm visualized. The patient was discharged on 4th post-operative day in stable condition.

Histopathological diagnosis of the tumor revealed undifferentiated malignant neoplasm. Microscopic findings showed: Grayish-brown soft to firm tissue shows highly cellular neoplasm arranged in sheets comprising moderately pleomorphic round-to-oval cells, coarse chromatin, moderate amount of cytoplasm, and indistinct nucleoli. Fair number of mitosis and necrosis is also shown. Perivascular arrangement of tumor cells is also shown.

After the HPE report, the patient and attendant were thoroughly explained and counseled about the condition and were referred to oncology center, where the patient was investigated thoroughly and underwent laparotomy with radical hysterectomy and received adjuvant chemotherapy.

DISCUSSION

All reported cases of uterine LMS associated with pregnancy were found incidentally in pregnancy. Kyodo *et al.* reported the only case in which they had observed the progress of the tumor during pregnancy.^[5] Similarly, the diagnosis of our patient was made histopathologically after operation. The fibroid was detected at the beginning of pregnancy and follow-ups with ultrasound examinations were performed. Neither any progress nor any clinically suspicious indications could be detected during pregnancy, and therefore, the tumor was mistakenly diagnosed as degenerating leiomyoma. As the fibroid was ruptured, with hemoperitoneum and tissues spread in the peritoneal cavity, attempt to remove maximum tissue with homeostasis, and drainage of hemoperitoneum during C/S was done. The indication for elective C/S at 31 weeks of pregnancy was

uncontrolled pain in abdomen and USG showing significant hemoperitoneum, which was alarming. Tumor growth during pregnancy is not unusual and has been demonstrated for uterine leiomyoma as a result of high estrogen and progesterone levels. In our patient, progression in size was documented during serial antenatal scans. The appropriate management of uterine LMS in pregnancy remains unclear. Since the patients are of a childbearing age, there is a desire to conserve fertility if possible. Uterine LMS is generally considered to be more aggressive than other types of uterine tumor and is well known for its frequent recurrence and resistance to chemotherapy. Therefore, an aggressive surgical procedure is usually considered to be the treatment of choice at the time of diagnosis.^[1] Our patient underwent a total abdominal hysterectomy with bilateral salpingo-oophorectomy, omentectomy, and paraaortal lymphadenectomy 6 weeks after CS. A total of 6 adjuvant chemotherapy cycles were given at an outside tertiary care center. The diagnosis of uterine LMS was made in the end third trimester and the pregnancy was not influenced by the disease. A case-control study in Iowa, USA, examined the effect of parity and age at first birth on the risk for various types of carcinomas treatment generally consists of surgical resection. In our patient, conservative management was possible for 3 days to allow fetal lung maturation, and

at the end of 31.3 weeks of gestation, an emergency C/S was performed.

In conclusion, we describe an extremely rare case of uterine leiomyosarcoma with rupture and hemoperitoneum in pregnancy. Treatment options seem to be reduced in pregnant women and mainly depend on the patient's condition as well as on the gestational age at presentation. In a multidisciplinary approach, an optimal therapy schedule should be assessed depending on the conditions.

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