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Case Report

Giant retroperitoneal liposarcoma during pregnancy: case report

Dhruba Prasad Paul, Kashish Garg*

Department of Obstetrics and Gynecology, Agartala Government Medical College and GBP Hospital, Agartala, Tripura, India

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***Correspondence:**

Dr. Kashish Garg,

E-mail: kashish201992@gmail.com

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ABSTRACT

24-year-old woman was presented with huge vulval mass for which she was operated, histopathological report of that mass shows angiomyoma. After few months she was discovered to have a giant retroperitoneal tumor incidentally during her routine obstetric examination at 24 weeks of gestation. Initial investigation by abdominal-pelvic computed tomography (CT) (18/08/18) revealed an a large multiseptated soft tissue attenuated minimally enhancing pelvic mass (31.8cmx13.2x 24cm) protruding in the abdominal cavity displacing the bowel loops proximally and pushing retro organs posteriorly. She underwent laparotomy with preservation of the fetus at 24 weeks of gestation. Final diagnosis was made after HPE report which shows myxoid liposarcoma. She was referred to regional cancer hospital for radiotherapy and then patient lost in follow up.

Keywords: Angiomyoma, HPE, Myxoid liposarcoma, Radiotherapy, Retroperitoneal tumor, Vulval mass

INTRODUCTION

In some cases, pregnancy can occur with large tumor masses in the abdomen. This compromises the pregnancy and asks for premature delivery or abortion, depending on when the tumor mass is diagnosed. The abdominal tumors which occur during pregnancy may be of gynecologic origin, other abdominal tumors, lymphomas or other undiagnosed abdominal masses. These masses need to be operated as soon as the pregnancy ends. Liposarcoma in pregnancy are peculiar clinical scenarios that are continuously being reported in the English literature for their rarity, challenges in management and speculations of association between pregnancy and disease kinetics. The World Health Organization classified liposarcomas into well-differentiated, myxoid, round-cell, pleomorphic and dedifferentiated subtypes; the first two subtypes belong to the low-grade lesions with better prognosis and higher incidence rates, which largely depends on the histologic subtypes, resection

margin and age of the patients.^{1,2} With free surgical margin, the 5-year overall survival rate can be 68-80%, although the local recurrence rate 75%.^{2,3} The peak prevalence of retroperitoneal liposarcoma (RPLS) is between the age of 40 to 70 years, usually seen in male 1.43:1. Liposarcoma in pregnancy is rare but deserves practitioners' attention, for early detection and proper surgical management could greatly improve disease outcomes.

CASE REPORT

A 24 years old women Mrs. X, residing at Ataharakant, Shantinagar Radhapur attended our hospital in the gynae OPD with complain of rapidly enlargement of vulva for last 6-7 months along with difficulty in passing stools and urine. After examination and discussion decision was made and patient was referred to surgery department. She was operated for the same huge vulval mass around 15x10cm in the month of March 2017.

Post op uneventful. Patient was discharged normally and advised to follow up monthly. Later on, biopsy report of that mass shows features suggestive of angiomyoma. In December 2017 she had again come with distention of abdomen for last 4 months with difficulty in breathing, walking, sleeping along with 5 months of amenorrhoea. She had history of 2 miscarriages, the cause of the recurrent miscarriage not known. Patient had menarche at the age of 14, after she had regular periods. Besides that, her past medical history was unremarkable. She did not remember her LMP exactly. During a routine obstetric examination, separate uterus not palpable as abdomen was hugely distended. Her body mass index (BMI) was 21.99 before this pregnancy when she presented to our hospital at 23 weeks pregnancy (by early week scan) her BMI was 30.09 with a good nutritional status. USG Shows (18/08/17)- A gestational sac around 6 weeks 6 days size seen with in endometrial cavity, yolk sac seen with in gestational sac, large multi-septed solid SOL with few hyperechoic and cystic lesion seen in peritoneal cavity.

The patient was then submitted to abdominal-pelvic computed tomography (CT) (18/08/17) revealed an a large multiseptated soft tissue attenuated minimally enhancing pelvic mass (31.8cmx13.2x 24cm) protruding in the abdominal cavity displacing the bowel loops proximally and pushing retro-organs posteriorly. However, there was no evidence of direct invasion to adjacent organ or metastasis. Laboratory findings showed mild leukocytosis and mild anaemia and other laboratory analysis revealed normal values of hepatic-renal function, urinalysis and electrolytes. Tumor marker studies showed increased level of CA-125 of 105.40 U/mL (normal range, 0 to 35), β -HCG-23,388 IU/ml. At the time of diagnosis, the size of tumor was very large (31x13x24 cm) and it was rapidly enlarging with high chance of being malignant tumor. It was determined that an early removal of the tumor will enhance the safety of the patient as well as the fetus. After a thorough discussion it was decided to perform the tumor removal along with continuation of pregnancy.



Figure 1: Huge vulval mass.

Intra-op findings

Abdomen opened through infra-umbilical vertical incision which was extended beyond umbilicus to assess the whole lump. Pregnant uterus was retracted anteriorly (24-26 weeks size). Both sides tubes and ovaries normal. Jelly like masses was found intraperitoneally which appeared to arise from intra-peritoneum around 15 kgs and masses sent for HPE. Hemostasis achieved and abdomen closed in layers. The patient had an uneventful recovery. The patient was discharged 15 days after surgical treatment without any immediate postoperative complication.

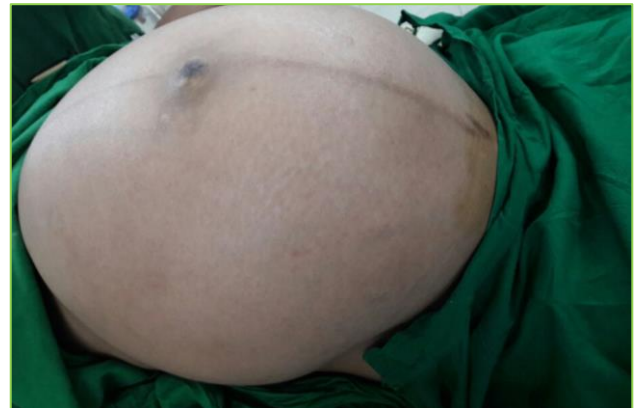


Figure 2: Abdomen before operation.

Later histopathological examination report shows Myxoid liposarcoma. The patient presented with pain abdomen after 1 month of operation followed by spontaneous expulsion of fetus. After that she had no problem, but the patient was referred to regional cancer hospital for radiotherapy and then patient lost in follow up.



Figure 3: Intra-op findings.

DISCUSSION

RPLS is one of the most common types of soft tissue sarcoma with peak prevalence between the age of 40 to

70 years, which is usually seen in male 1.43:1.^{4,5} RPLS during pregnancy is extremely rare and a challenge for both the surgeon and pregnant woman.⁶

Liposarcoma is categorized into five histological subtypes: well-differentiated, myxoid, round-cell, pleomorphic, and dedifferentiated (WHO classification, 2002). Well-differentiated and myxoid subtypes belong to the low-grade lesions, whereas the other subtypes are considered as high-grade lesions.

Myxoid liposarcoma is the second most common subtype, consists of 18% of all liposarcoma, and is the most common subtype in young patients.⁷ It is often found with painless palpable mass arising in the thigh and retroperitoneal space, especially for young patients.⁴ MRI is a highly reliable radiological method in diagnosing these tumors, especially for assessment of pregnant woman. With careful interpretation, the MRI can provide us with enough information for accurate diagnosis differentiation and even identification of subtypes.⁸⁻¹⁰ Myxoid liposarcoma has a relatively characteristic appearance as a well-circumscribed multinodular mass with low signal intensity on T₁WI and a high but heterogeneous intensity on T₂WI.

The relationship between pregnancy and liposarcoma is not clear. Cantin and Mc Neer considered that pregnancy does not adversely affect the prognosis of the tumor. Whereas, they suggested that an estrogen-progesterone environment possibly has a favorable impact on the natural history of sarcoma, and hormone therapy in the management of metastatic sarcoma should be explored.¹¹

The retroperitoneal space is rather vast; RPLS has no specific symptoms in the early stage. Although surgery is the mainstay of the therapeutic modalities, the RPLS is usually diagnosed in the advanced stage with a palpable mass, and contiguous organ resection is often indicated.¹² If resected with negative surgical margin, the 5-year overall survival rate can be 68% to 80%, although local recurrence rate can be as high as 75%¹². The extended surgery, contiguous organ resection, major vessel resection, and reconstruction are often indicated to get a negative surgical margin and a better prognosis.¹³

This patient was diagnosed with giant retroperitoneal tumor incidentally during her routine obstetric examination. The therapeutic protocol is challenging because: 1) The patient has a firm willing of preserving the fetus. 2) The resection is urgent as the tumor is expanding. 3) The impact on the fetus should be assessed such as time of gestation, hemorrhage, length of operation, medications and anesthesia.

Authors reviewed the literature; only 16 cases of liposarcoma during pregnancy were reported in English literature, ten were derived from retroperitoneal space, and five cases are myxoid subtype. Surgery was performed in five cases after delivery, five cases

concurrent with cesarean section. Three patients died within the first year after surgery.¹⁴⁻¹⁷ In only two cases, surgeries were performed during pregnancy in 13 weeks of gestation.^{18,19} Most surgery were performed postpartum or at the time of cesarean section.

CONCLUSION

In rare cases, pregnancy can be compromised by large intra-abdominal tumors which can put the health of both the mother and the fetus in danger. It's suggested that future mothers are regularly examined, prior to pregnancy, so that they can keep track of their health and ability to reproduce. The entire pregnancy period should be carefully observed, too, since the symptoms can sometimes lead to interdisciplinary examinations and consultations with other specialists. This will ensure mothers are healthy and can give birth to healthy offspring.

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