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Case Report

Spontaneous rupture of leiomyosarcoma

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ABSTRACT

Uterine sarcomas are rare, accounting for approximately 3-7% of uterine malignancies. Leiomyosarcoma (LMS), a particularly aggressive subtype, often presents diagnostic challenges due to overlapping features with benign fibroids. Spontaneous rupture of LMS is exceptionally rare and may lead to life-threatening complications. A 42-year-old woman presented with severe lower abdominal pain, distension, dyspnea, and hemodynamic instability. Imaging revealed a ruptured uterine mass with hemoperitoneum, bilateral pulmonary embolism (PE), and suspected metastatic disease. Initial stabilization required ICU support and multidisciplinary input. Uterine artery embolization was performed to control haemorrhage. The patient developed subsequent infections but stabilized and was repatriated to her home country, where she received six cycles of chemotherapy. Surgery is planned following completion of chemotherapy. This case underscores the diagnostic difficulty in differentiating LMS from benign fibroids, especially in the presence of large, rapidly growing uterine masses. Delayed diagnosis can result in catastrophic events such as spontaneous rupture. Imaging and histology remain essential, but molecular profiling is increasingly valuable for therapeutic planning. Spontaneous rupture of LMS, though rare, should be considered in patients with large symptomatic uterine masses. Early recognition, multidisciplinary management, and genetic profiling are vital for optimizing outcomes. Proactive surgical management of large tumours may help prevent such complications, even in presumed benign cases.

Keywords: Uterine leiomyosarcoma, Spontaneous tumour rupture, Hemoperitoneum, Advanced uterine sarcoma

INTRODUCTION

Uterine sarcomas are a rare group of tumours that comprise about 3-7% of uterine malignancies. Subtypes include LMS, adenosarcoma, endometrial stromal sarcoma, and undifferentiated sarcoma. Some uterine sarcomas are not easily classifiable, including those with a histologic appearance similar to fibrosarcoma.[1-4] This article aims to raise awareness about rare uterine sarcomas and one of their serious complications: spontaneous rupture.

CASE REPORT

A 42-year-old widow (parity 2) was admitted to our hospital on November 7, 2023, with severe, constant lower abdominal distension and pain, low-grade fever, and difficulty breathing. Her normal menstrual cycle lasted 5–6 days every 30 days, but she had experienced heavier and prolonged bleeding associated with moderate to severe abdominal pain over the past few years. However, she denied intermenstrual bleeding.

On presentation, the patient appeared mildly distressed, with moderate pallor. A pelvic examination revealed a tender, twenty-week-size pelvic mass and a distended abdomen. A CT pulmonary angiogram (CTPA) showed extensive bilateral pulmonary embolism (PE), left proximal deep vein thrombosis (DVT), bilateral pulmonary nodules, and possible metastatic uterine malignancy with lung and bone metastases. A pelvic scan also revealed discontinuity at the anterolateral aspect of the mass, suggesting rupture, along with a large-volume hemoperitoneum. She required oxygen supplementation to maintain saturation and had the unstable vital signs (Figure 1).

Active bleeding from the tumour was evident due to a rapid decline in haemoglobin levels. Her condition necessitated intensive care unit (ICU) monitoring with inotropic support. An urgent multidisciplinary team discussion involving a gynaecologist, oncologist, interventional radiologist, and the PE response team (PERT) recommended a palliative approach, considering her guarded prognosis and the advanced oncological disease.

The team concluded that she was not a suitable candidate for thrombectomy or extracorporeal membrane oxygenation (ECMO) and opted for conservative management without surgical intervention.

After stabilizing her haemoglobin with blood transfusions, a low dose of low-molecular-weight heparin (clexane) was initiated to manage the PE, with the intention of gradual up titration to a therapeutic dose as tolerated. A few days later, she developed sudden-onset abdominal pain, a tense abdomen, and a drop in haemoglobin, suggesting ongoing bleeding. An urgent image-guided uterine artery embolization was performed to arrest tumour bleeding, followed by abdominal drain insertion to remove ascitic fluid.

She subsequently developed a urinary tract infection, followed by hospital-acquired pneumonia, requiring parenteral tazocin. Over the next few days, her condition stabilized, as well as she was transported back to the her home country with the medical escort service, as per her request.

In her hometown, she received six cycles of chemotherapy (Paclitaxel/carboplatin), and a repeat CT scan showed residual pulmonary metastases. The multidisciplinary team decided to complete six cycles of chemotherapy and planned for surgery. The patient experienced disease recurrence and pulmonary metastases following six cycles of chemotherapy and was planned for the second-line treatment. Unfortunately, her condition deteriorated, and she passed away seven months after the initial diagnosis.

She had previously visited our hospital in 2021 for heavy menstrual bleeding (HMB) and a palpable abdominal lump during a routine medical checkup. A routine scan at that time revealed: Uterus size (cm): 13.9×12.2×6.4, endometrium thickness (mm): 2.0, findings: fluid within the myometrium; well-circumscribed masses likely due to fibroids, largest fibroid size (cm): 11.0×8.3×6.8 (IM/LW) with a mass effect on the endometrium, right ovary size (cm): 1.9×1.6×1.2 (volume: 1.9 cm^3)-unremarkable, left ovary size (cm): 3.5×2.3×1.5 (volume: 6.3 cm^3)-unremarkable and adnexa/POD: No significant adnexal mass or fluid detected.

Due to the large fibroid, surgical management was offered, but she decided to undergo the procedure in her hometown and did not return for the further follow-up (Figure 2 and 3).

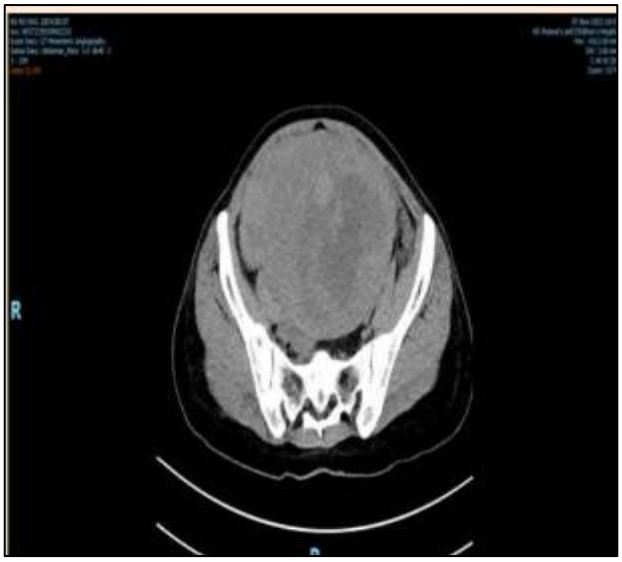

Figure 1: CT mesenteric angiography of huge fibro myosarcoma and hemoperitoneum.

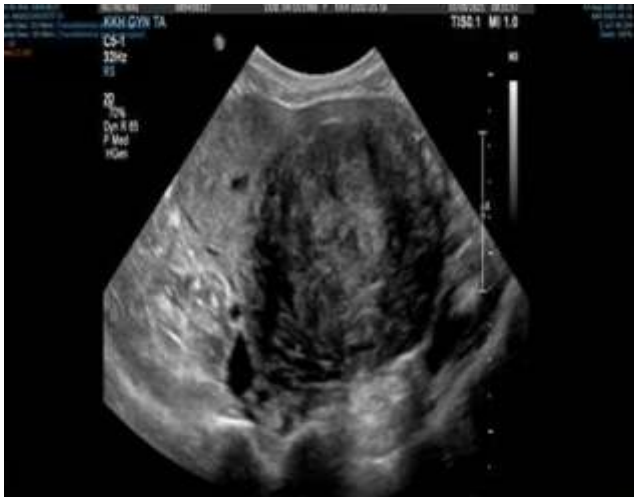

Figure 2: Huge uterine fibroid in 2021.

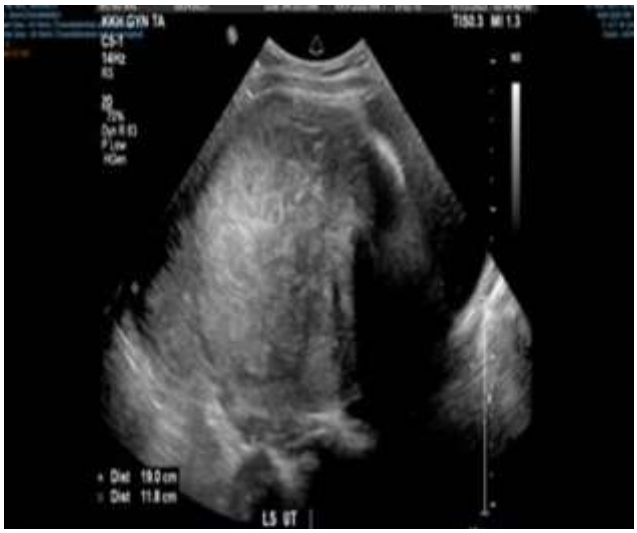

Figure 3: Uterine fibro-myosarcoma in 2023.

Table 1: Uterine sarcoma staging system by FIGO.[7]

Stages	Description	
	Leiomyosarcomas/ESS	**Adenosarcoma**
I	Limited to the uterus	
IA	Tumour ≤5 cm	Limited to the endometrium and/or endocervix
IB	Tumour >5 cm	Invading <50% myometrium
IC	-	Invading >50% myometrium
II	Extending beyond the uterus but within the pelvis	
IIA	Involving adnexa	
IIB	Involving other pelvic tissues	
III	Infiltrating abdominal tissues	
IIIA	1 site	
IIIB	>1 site	
IIIC	Regional lymph nodes metastases	
IVA	Invading bladder or rectum	
IVB	Distant metastases	

DISCUSSION

The diagnosis of uterine sarcomas can be challenging, as demonstrated in this case. Extensive sampling and immunohistochemical analysis of LMS patients revealed a spindle cell neoplasm with mild to moderate nuclear atypia, focal tumour cell necrosis, and increased mitotic activity. Smooth muscle markers yielded ambiguous results, with some markers being negative (desmin and caldesmon) and others showing focal positivity (smooth muscle actin). Given the clinical presentation, gross examination, and histologic evaluation at the time of diagnosis, the diagnosis of "most consistent with LMS " was made, and they were treated accordingly.

In retrospect, molecular testing would have been valuable in this case. Unfortunately, the patient's initial treatments were unsuccessful. Regarding targeted therapy, if the patient's tumour was found to have a COL1A1-PDGFB gene fusion (collagen type I alpha 1 gene with the platelet-derived growth factor beta chain), which is most commonly associated with dermatofibrosarcoma protuberans (DFSP) and may be treated with targeted therapy.[5]

Ultrasound examination is the first step in evaluating uterine tumours; however, a limited field of view can be a challenge with large tumours. Certain sonographic features, such as tumour size >5 cm, high vascularization, irregular borders, and necrotic areas, are more commonly associated with LMS and smooth muscle tumours of uncertain malignant potential (STUMP). The morphological uterine sonographic assessment (MUSA) has been proposed by consensus to describe the ultrasonographic characteristics of the myometrium and uterine masses.

According to MUSA, STUMP exhibits greater colour Doppler enhancement than benign masses due to increased intralesional and perilesional vascularization.[6]

There are no definitive clinical or radiological criteria to distinguish uterine sarcomas from benign uterine leiomyomas. Both conditions commonly present with abnormal vaginal bleeding, pelvic pain, and an abdominal mass. However, a rapidly growing uterine fibroid in a perimenopausal or postmenopausal woman should raise suspicion of sarcoma. The initial diagnostic approach typically involves a pelvic ultrasound, followed by pelvic magnetic resonance imaging (MRI).[7]

LMS and undifferentiated sarcoma

Stages I and II

Most women undergo surgery to remove the uterus (hysterectomy) along with the fallopian tubes and ovaries (bilateral salpingo-oophorectomy). However, in women who are still having regular menstrual cycles, the ovaries may not be removed. Pelvic and para-aortic lymph node dissection or laparoscopic lymph node sampling may be performed if swollen nodes are detected on imaging tests or felt during the operation.

During surgery, nearby organs and the peritoneum (the thin membrane lining the pelvic and abdominal cavities) are examined to determine if the cancer has spread beyond the uterus.[7]

Some stage I cancers may not require further treatment after surgery, and observation (close monitoring post-surgery) is an option. In other cases, adjuvant treatment-radiation therapy, with or without chemotherapy-may be necessary if there is a high risk of cancer recurrence.

The goal of surgery is curative intent with R0 resection (complete tumour removal with no residual disease), but the surgeon can only remove what is visible. Microscopic cancer cells that are too small to detect may remain. Post-surgical treatments can help eliminate these residual cells to reduce the risk of recurrence.

For LMS of the uterus, adjuvant radiation therapy may decrease the risk of local recurrence (cancer regrowth in the pelvis), but it does not appear to improve overall survival outcomes.[7] Since LMS can recur in distant organs, such as the lungs, some experts recommend adjuvant chemotherapy after surgery for stage II cancers. Chemotherapy is sometimes suggested for stage I LMS, but its benefits remain uncertain. Studies on adjuvant chemotherapy for early-stage LMS have shown promising results in reducing recurrence, but they have not yet demonstrated a clear survival benefit. Ongoing studies continue to investigate the effectiveness of adjuvant treatments.[7]

Leiomyosarcoma and undifferentiated sarcoma stage III

Surgery is performed when the surgeon determines that all of the cancer can be removed. This typically includes hysterectomy (removal of the uterus), bilateral salpingo-oophorectomy (removal of both fallopian tubes and ovaries), resection of any other organs involved with the tumour, and lymph node dissection or sampling. If the tumour has spread to the vagina, partial or complete vaginectomy (removal of the vagina) may be necessary.

After surgery, radiation therapy or chemotherapy may be recommended to reduce the risk of disease recurrence. Patients with multiple comorbidities and a poor ECOG (Eastern cooperative oncology group) performance status, who have been assessed as unfit for surgery, may be offered chemotherapy, radiation therapy, and/or chemoradiation as alternative treatment options.[7]

Stage IV

Stage IV cancers have spread to nearby organs and tissues, such as the bladder or rectum. Surgical resection may be considered in selected cases. If surgery is not feasible, radiation therapy may be administered either alone or followed by chemotherapy.

Stage IVB cancers have spread beyond the pelvis, most commonly to the lungs, liver, or bones. There is no standard treatment for these cancers. Chemotherapy may help shrink the tumours temporarily. Radiation therapy, followed by chemotherapy, may also be an option. In cases where other treatments are ineffective, targeted drug therapy or immunotherapy may be considered.[7]

CONCLUSION

Spontaneous rupture of LMS, though rare, should be considered in patients with large symptomatic uterine masses. Early recognition, multidisciplinary management, and genetic profiling are vital for optimizing outcomes. Proactive surgical management of large tumours may help prevent such complications, even in presumed benign cases.

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