Case Report

Atypical presentation of septic fibroid as uterine perforation

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ABSTRACT

Pyomyoma, also known as suppurative leiomyoma, is a rare complication of uterine leiomyoma which arises due to infarction, degeneration, and subsequent infection of a pre-existing leiomyoma. This report highlights the case of a 52-year-old patient who presented to the emergency department with complaints of fever, abdominal pain, and breathlessness. She was a known case of fibroid uterus. She also had uncontrolled diabetes mellitus and bilateral renal calculi. The patient was diagnosed to have diabetic ketoacidosis with shock and was admitted to the intensive care unit. Computed tomography scan showed incidental findings of rent in the fundus of the uterus suggestive of uterine perforation. After stabilization of the patient, laparotomy was done which revealed the uterus to be enlarged to 14 weeks size. There was a 3 cm × 3 cm perforation on the fundoposterior surface of the uterus with purulent discharge. A decision was taken for total abdominal hysterectomy. The patient had a remarkable improvement postoperatively.

Key words: Diabetes ketoacidosis, Pyomyoma, Uterine rupture

Fibroids are among the most common benign neoplasia occurring in about 70% of women [1]. In 25% of women of reproductive age, they are symptomatic and require treatment [1]. Pyomyoma is a rare but life-threatening condition that results from the infarction and infection of uterine leiomyoma [2]. Clinicians should consider the diagnosis of pyomyoma in women with leiomyoma and sepsis in the absence of any apparent source of infection. While there have been around 100 cases of pyomyomas reported in the literature, we present this case to highlight its atypical presentation as rupture uterus.

CASE REPORT

A 52-year-old multiparous lady was brought to the emergency department with complaints of high-grade fever, breathlessness on routine activities, and lower abdominal pain for the past 3 days. The pain was insidious in onset, dull aching type, not responded to over-the-counter analgesics, and in fact, increased over the past 3 days. On further inquiry, the patient gave a history of diabetes mellitus for the past 10 years. She had been on insulin, however, was non-compliant with the treatment. She had been diagnosed to have a large 6 cm × 5 cm intramural fibroid on the posterior wall of the uterus when she presented with heavy menstrual bleeding 6 months back and had been started on oral progesterone (Tablet Norethisterone 5 mg bd) 3 months back. The patient had been planned for a hysterectomy after achieving glycemic control. She was also a known case of bilateral renal calculi, had undergone right percutaneous nephrolithotomy 2 months back at a tertiary care center.

On examination, the patient appeared toxic and was tachypneic. Her pulse was 120/min feeble, blood pressure was 70 mmHg systolic, respiratory rate was 40/min with no added sounds, and the temperature was 101°F. On abdominal examination, there was a palpable mass, arising from the pelvis and there was tenderness on deep palpation. No guarding, rigidity, and shifting dullness were found on abdominal palpation. Liver, spleen, and kidney were not enlarged. There was no renal angle tenderness. On per vaginal examination, the uterus was irregularly enlarged to 14 weeks; there was tenderness over the uterus and in the posterior fornix. The cervix was closed. No obvious forniceal mass was felt.

Random blood sugar at admission was 450 mg%. Urine showed heavy ketonuria. She was admitted to the intensive care unit as a case of diabetic ketoacidosis and was managed with intravenous fluid, insulin infusion, inotropes, and high-end antibiotics (Inj. Meropenem 500 mg iv 8 hourly, Inj. Aztreonam 1g iv 8 hourly, and Inj. Clindamycin 600 mg iv 8 hourly).

Computed tomography (CT) scan abdomen was done which showed bilateral hydronephrosis. The surprising CT findings were the presence of multiple air foci within the endometrial cavity. These air foci were seen to reach up to the serosal surface of the

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fundus with communication with the extraserosal air pocket. There was also a rent in the fundus measuring 6 mm suggestive of uterine perforation (Fig. 1). The patient did not give a history of dilatation and curettage.

The patient improved in the intensive care unit and was gradually weaned off from the inotropes. The urologists decided on a conservative line of management for the hydronephrosis. We decided to follow the dictum for the management of sepsis – elimination of the source of sepsis. After adequate counseling of the patient and relatives for the need of surgery, a decision for laparotomy was taken after 1 week of admission despite the comorbidities (diabetes and bilateral hydronephrosis).

With the patient under spinal anesthesia, the abdomen was opened with a transverse incision. The operative findings were: The uterus was found to be enlarged to 14 weeks size. There was a 3 cm × 3 cm perforation on the fundoposterior surface of the uterus, with the sigmoid colon, transverse colon, and loops of small intestine adherent to the site (Fig. 2). The bowel loops at the site of adhesion looked inflamed. There were also superficial serosal tears on the mesentery of the large colon. During careful separation of the adhesions, around 50 mL pus was drained, which was sent for culture sensitivity studies. A decision was taken to proceed with total abdominal hysterectomy with bilateral salpingo-oophorectomy. Surgeons were also called to help separate the adhesions and confirm the integrity of the bowel and mesentery. The pelvic drain was kept in situ, before closing the abdomen.

The patient had a marked clinical improvement postoperatively. Culture sensitivity reports showed growth of *Enterococcus faecalis*, which was sensitive to linezolid. The drain was removed on the 7th post-operative day. The patient was discharged on the 14th post-operative day. Subsequent histopathological analysis of the uterus revealed a leiomyoma with acute suppurative inflammation, congestion, necrosis, and rupture, thus collaborating the clinical suspicion of pyomyoma.

DISCUSSION

Since its first description in 1871, less than 100 cases of pyomyomas have been reported [3]. The most likely cause of pyomyoma is a vascular compromise, followed by bacterial seeding of necrotic foci within the leiomyoma from direct, hematogenous, or lymphatic spread [2]. Pregnant women, postpartum, or post-abortion women, those with a history of instrumentation of the uterus, post-menopausal, and immunocompromised women are specifically vulnerable to myomatous infections [4]. In the postpartum period, women are at risk of myoma infection because of the increased risk of hemorrhage or ischemia of the fibroid resulting from hormonal changes [4]. Other factors closely linked with pyomyomas pre-existing uterine or cervical infections [5]. Interestingly, the increased incidence of pyomyomas in recent years has coincided with the increased use of uterine artery embolization as a modality of treatment for fibroids [6].

Pyomyoma is more common in submucosal leiomyomas because their supply is relatively tenuous and their position adjacent to the uterine lumen predisposes them to ascend infection [7]. The interval between the initial onset of symptoms and diagnosis varies greatly as pyomyoma may present itself with abrupt onset or may extend to a year of incubation with progressive dissemination of the infection [4]. A typical triad of its clinical features is the presence of fibroid, sepsis, and absence of any other source of infection [6]. The infected leiomyoma can manifest as a painful abdominal or pelvic mass, as bacteremia without a clear focus of infection, or as an acute abdomen due to intra-abdominal rupture [6].

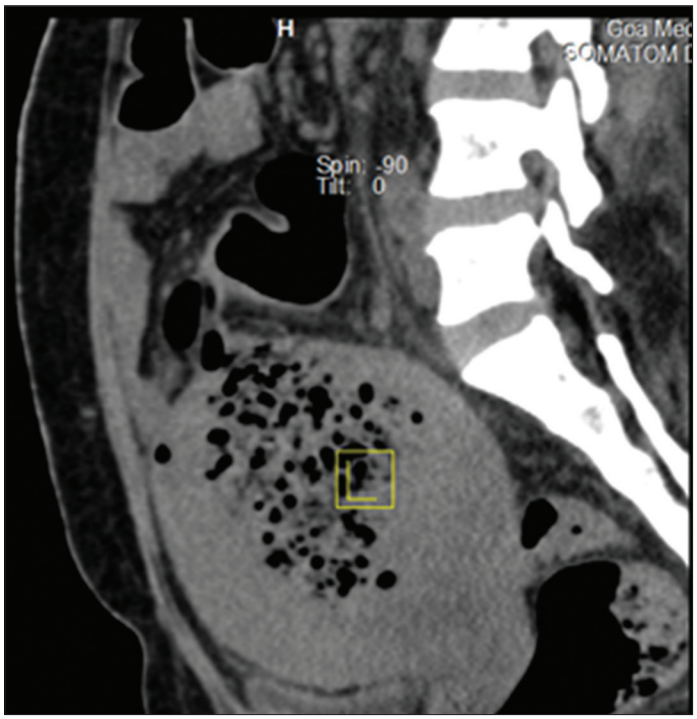

Figure 1: Contrast-enhanced computed tomography section showing uterine perforation with gas in the uterine cavity

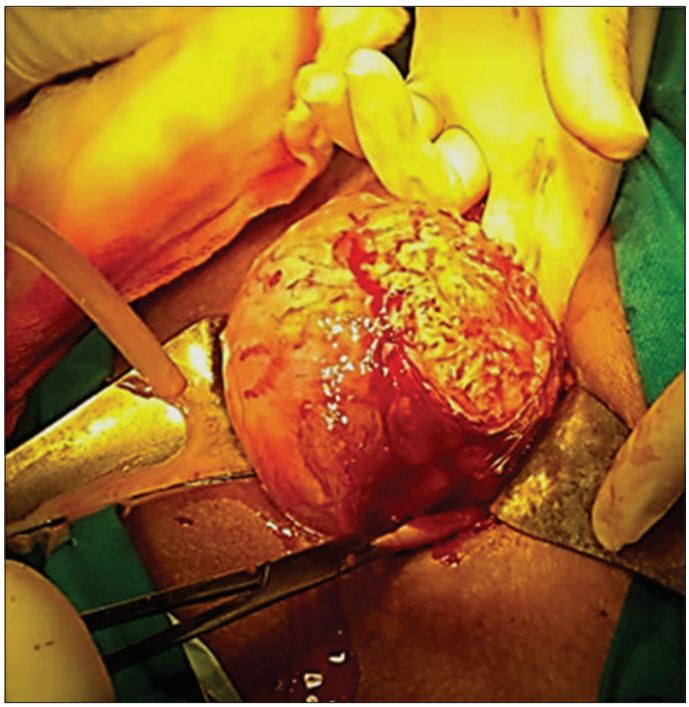

Figure 2: Perforation noted on the fundoposterior aspect of the uterus

Early diagnosis and treatment are the cornerstones in the management of this condition [6]. The diagnosis of pyomyoma is difficult because of its diverse presentation and lack of characteristic imaging features [8]. The ultrasonography findings in this condition include heterogeneous echogenic pelvic mass with mixed solid and cystic components, reverberation artifacts, acoustic shadowing indicating the presence of gas, and discontinuity of the uterine wall [6]. CT findings in suppurative leiomyomas include the presence of gas and debris in the leiomyoma, heterogeneous appearance, and thickened fibroid wall [6]. Ruptured pyomyomas are distinguished by intraperitoneal free air, ascites, and discontinuity of the uterine wall [6]. Histopathology may show cystic degeneration, hyaline change, hemorrhage, necrosis, and acute inflammatory changes with smooth muscle cells [9].

The differential diagnosis for this condition includes endometritis with cervical obstruction, ruptured or tubo-ovarian abscess, aseptic necrobiosis, and bowel invasion from a gynecologic malignancy leiomyosarcoma, or ovarian cancer [9]. Delay in diagnosis may have severe consequences such as rupture with peritonitis, renal cortical necrosis, deep vein thrombosis by compression, endocarditis, pancreatitis, and death [4].

Organisms reported to cause pyomyomas include *Clostridium*, *Staphylococcus aureus*, *Streptococcus milleri*, *Streptococcus hemolyticus*, *Proteus*, *Serratia marcescens*, *Actinomyces meyeri*, *E. faecalis,* and *Staphylococcus agalactiae* [5]. Rare cases of infection by *Sphingomonas paucimobilis*, an opportunistic pathogen emergent in the nosocomial setting, have been reported [10].

The definitive treatment is hysterectomy, however, there have been case reports of nulliparous patients managed conservatively by myomectomy [4]. Recent advances in laparoscopic techniques now allow for the use of minimally invasive approaches and, more specifically, the avoidance of laparotomy with in-bag morcellation [11]. Without appropriate treatment, mortality rates as high as 20% have been reported [3].

CONCLUSION

While there have been a few reports of pyomyomas in the literature, its presentation as rupture uterus is exceedingly rare. We present this case to highlight the need for good clinical acumen and sound judgment to achieve a good patient outcome even in this era of advanced technology. Gynecologists should consider the diagnosis of pyomyoma in women with leiomyoma and sepsis in the absence of any apparent source of infection.

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