

Double Trouble Leiomyoma- A Twisted Leiomyoma as a Cause of an Obstructive Acute Pyelonephritis

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ABSTRACT

A 57-year-old woman presented to the emergency department with a clinical condition suggestive of renal colic. The image exams showed several non-obstructive right kidney stones, a discrete right kidney ectasia and a poliomyomatous uterus. She was discharged after improvement with analgesia. Four days later, the woman presented with strong right iliac fossa pain, nausea and diminished bowel activity. She had signs of peritoneal irritation and the CT scan demonstrated right moderate uretero-hydronephrosis related to pelvic ureter compression by uterine leiomyomas. The patient was treated with right retrograde ureteral stent placement and endovenous antibiotics with improvement. In the follow-up period, the decision was to perform an abdominal hysterectomy due to the presence of an enlarged poliomyomatous uterus, with a subserous posterior polylobulated myoma with 100*72*52 mm. The surgical exploitation showed two posterior subserous myomas with a twisted common pedicle and a necrotic appearance. The post-surgical period was uneventful.

Keywords: leiomyoma, Torsion, Pyelonephritis

INTRODUCTION

Uterine leiomyomas are among the most common pathologies of the female urogenital tract, affecting up to 70% of the female population [1]. Most often asymptomatic, they can cause symptoms in about 30% of women, depending on the location and size. The most common symptom is abnormal uterine bleeding, which can lead to anemia and fatigue. By compression of the surrounding organs, fibroids can also result in pelvic/abdominal pain, urinary or gastrointestinal symptoms or even hydronephrosis. They are also associated with infertility and adverse obstetric outcomes [2]. Uncommonly, leiomyomas can provoke acute complications including torsion of subserous pedunculated myoma, acute urinary retention, acute pain by red degeneration and acute intra-peritoneal hemorrhage [3]. Although rare, these acute complications can cause serious morbidity and have a significant impact on woman's quality of life.

The management of leiomyomas depends on the symptoms, number, size and location of the masses and woman's desire to preserve fertility. Medical treatment is the primary option for leiomyoma-associated symptoms. Surgical treatments include conservative (myomectomy) and radical (hysterectomy) approaches [2].

The authors describe a case of a twisted pedunculated uterine leiomyoma causing necrosis and peritonitis followed by uretero-hydronephrosis by compression. For the elaboration of this article, an informed consent was obtained from the patient.

Case Report

A nulliparous, postmenopausal 57-year-old female presented to the emergency department with right lower back pain with irradiation to ipsilateral iliac fossa. She had a medical history of obesity, hypertension, dyslipidemia, type 2 diabetes and renal lithiasis. At physical examination, there were no signs of peritoneal irritation and the Murphy sign was positive at the right side. The blood test demonstrated a normal kidney function, no leukocytosis and normal C-reactive protein level. A computed tomography (CT) scan showed multiple non-obstructive right kidney stones and a discrete right kidney ectasia with no evident obstructive ureteral stones. The uterus had several round masses between 3 and 7 cm suggestive of leiomyomas [Figure 1]. A gynecological evaluation was requested and there was no abnormal leukorrhoea and the uterus was not painful at mobilization. The transvaginal ultrasound revealed a retroverted uterus with several masses suggestive of fibroids, the largest of which was a conglomerate of three subserous leiomyomas with 73 mm on the posterior uterine wall. There was also a small

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amount of free fluid in the pouch of Douglas. Due to suspicion of renal colic and improvement with analgesia, the woman was prescribed analgesic medication and discharged.

Four days later, the patient presented again to the emergency department with strong right iliac fossa pain and nausea. She also referred diminished bowel activity. She was afebrile and the Murphy sign was negative bilaterally. The abdominal palpation showed rebound tenderness on the right iliac fossa. She had leukocytosis and elevated C-reactive protein level. A new CT scan demonstrated right moderate uretero-hydronephrosis, probably related to pelvic ureter compression by partially calcified uterine leiomyomas [Figure 2]. The patient was hospitalized due to an obstructive acute pyelonephritis, submitted to right retrograde ureteral stent placement and treated with endovenous antibiotics. She improved clinically and analytically and was discharged at day 9. The urine cultures were negative.

An abdomino-pelvic magnetic resonance image performed one month later demonstrated an enlarged poliomyomatous uterus,

with emphasis on a subserous, well delimited but polylobulated myoma on the posterior uterine wall with 100*72*52 mm and heterogenous intensity signal on T2- weighted images [Figure 3]. After discussion with the patient about the therapeutic options and prognosis, the decision was to perform an abdominal hysterectomy with bilateral salpingo-oophorectomy. The surgical exploitation showed epiploon and colon adhesions to the posterior uterine wall. After adhesiolysis, we verified a poliomyomatous uterus with 8 cm and a 10 cm posterior mass constituted by two subserous myomas with a common pedicle. The pedicle was twisted and the myomas had a necrotic appearance [Figure 4, 5]. After extraction of the uterus, we verified inflammatory signs in the pouch of Douglas. The procedure was completed without further events. The post-surgical period was uneventful and the patient was discharged at day 3.

The histological examination revealed multiple intramural and subserous myomas with large areas of ischemic necrosis and regenerative changes. It also demonstrated an endometrial polyp with hyperplasia with atypia.

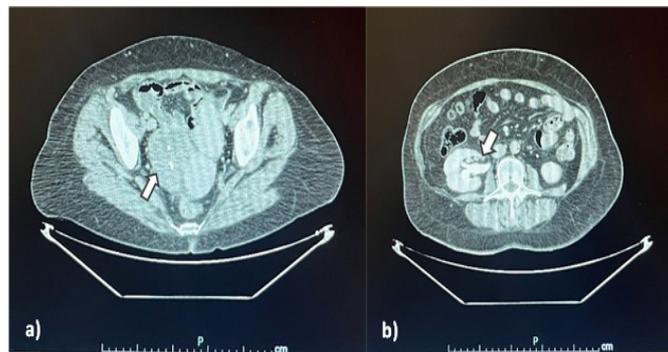


Figure 1: CT scan performed the first time the patient presented to the emergency room. a) The uterus had several masses suggestive of leiomyomas (arrow). b) The scan demonstrated a discrete right kidney ectasia (arrow).



Figure 2: CT scan performed the second time the patient presented to the emergency room, four days later. It demonstrated right moderate uretero-hydronephrosis (arrow), probably related to pelvic ureter compression by partially calcified uterine leiomyomas.



Figure 3: Abdomino-pelvic magnetic resonance image performed one month later from the acute presentation. The T2- weighted image showed a well delimited but polylobulated myoma on the posterior uterine wall with heterogenous intensity signal.

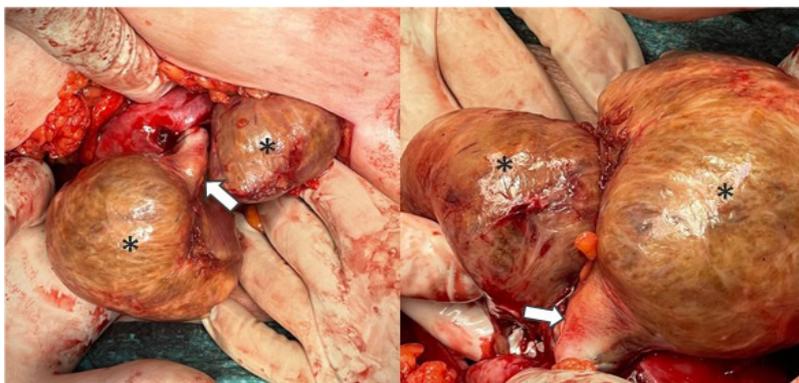


Figure 4: Surgical exploitation showed a poliomyomatous uterus and a 10 cm posterior mass constituted by two subserous myomas (asterisk) with a common pedicle (arrow) was twisted and the myomas had a necrotic appearance.



Figure 5: The two posterior myomas with a common pedicle after surgical removal. The maximum diameter of the mass was 10 cm.

DISCUSSION

Uterine leiomyomas are the most common pelvic tumors in women and the most frequent primary indication for hysterectomy in Portugal [4].

On the other hand, torsion of a pedunculated leiomyoma is a rare acute surgical emergency with a reported incidence of less than 0, 25% in patients with subserous uterine leiomyomas undergoing surgical intervention [5]. Torsion of a subserosal myoma depends on three factors [6, 7]:

- i) The presence of a long and thin pedicle in order to allow rotation and torsion of the myoma;
- ii) Larger and heavier fibroids are more prone to twist on themselves. On the other hand, they are less likely to untwist;
- iii) The relationship between the leiomyoma and adjacent viscera.

The symptoms are variable, depending on the speed at which the torsion occurs and on its degree of rotation. With partial torsion with spontaneous untwisting, the complaints may solve promptly. Conversely, a completely twisted pedicle initially produces venous stasis with oedema and this, in turn, eventually compresses the arterial blood supply causing hemorrhagic necrosis and gangrene [6]. If left untreated, hemorrhagic infarction of the involved myoma may be followed by infection and peritonitis [6, 8]. Associated hemoperitoneum and even hypovolemic shock have also been reported [9, 10]. Moreover, Guglielmo et al. described a case of small bowel volvulus due to torsion of pedunculated fibroid [11].

The accurate diagnosis of torsion of a pedunculated myoma is challenging. The clinical signs and symptoms including abdominal pain, gastrointestinal disturbances, pelvic mass and peritoneal

irritation are not specific. Thus, initial clinical presentation can be misleading and similar to other causes of acute abdomen such as acute appendicitis, pelvic inflammatory disease, adnexal torsion and red degeneration of fibroid. In our case, the diagnosis was even more difficult due to presence of other confounding factors such as the right uretero-hydronephrosis, the presence of renal lithiasis and the initial clinical presentation with right lower back pain and the positive Murphy sign. Albeit logically possible, severe obstructive uropathy caused by compression of a uterine myoma is rare. There are sporadic cases reported in the literature of uterine leiomyoma associated with hydronephrosis, acute pyelonephritis and even impaired renal function [12, 13, 14].

Ultrasound, CT and magnetic resonance imaging (MRI) have been used for diagnosis of leiomyoma torsion but the findings are inconsistent and not yet well established [15, 16]. Ultrasound should be the primary imaging modality of choice for study of acute pelvic pain and suspected gynecological diseases. It can detect a subserous fibroid and may identify a torsion when colour Doppler demonstrates a twisted pedicle or when there is gap in vascularization between the myoma and the uterus. However, especially when the pedicle is thin and invisible, ultrasound can be ineffective [17]. In an emergency department setting, CT is commonly used as the first examination to clarify a strong, acute abdominal pain if a gynecologic disorder is not initially suspected. Roy et al. [8] proposed a diagnosis of acute torsion of subserous leiomyoma using CT when normal ovaries and contrast enhancement of the uterine portion connected to the mass are present. Other reported typical CT features associated with subserous leiomyoma torsion include poor contrast enhancement inside the myoma, thin rim enhancement around it and a dark fan sign (fan-shaped, poor contrast enhancement in the uterus area adjacent to the myoma)

[8,17,18]. MRI is a non-invasive and accurate image modality for detection and characterization of uterine leiomyomas. It is the exam of choice to complement the findings of ultrasound. In the case of necrobiotic myomas, the mass has a heterogenous hyper-intense T2 signal, and hyper or iso-intense T1 signal depending on hemorrhagic or ischemic mechanism. It may also be present a peripheral halo, hypo-intense on T2 and hyper-intense on T1 that corresponds to venous outflow obstruction [16].

Nevertheless, surgery with direct visualization remains the primary method of diagnosis and treatment of a twisted myoma [5, 9, 15]. Depending on the patient and clinical situation, the surgical choice can be a myomectomy or a hysterectomy. Judicious intervention is essential as delayed diagnosis and management are associated with increased morbidity and even mortality [9].

In conclusion, torsion of a pedunculated subserous leiomyoma is a gynecological emergency whose diagnosis implies a high index of suspicion. Although clinical presentation and image exams can raise the suspicion, definite diagnosis is mainly established during surgery. Timely diagnosis and prompt intervention are essential to avoid life-threatening consequences such as sepsis or internal bleeding with hypovolemic shock.

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