Torsion of a uterine leiomyoma – a rare cause of hemoperitoneum; a case report and review of the literature

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Abstract

Uterine leiomyomas are common benign uterine tumors but by contrast, their acute complications are very rare. We present an unusual case of 38-year-old woman that came to the emergency department with acute abdominal pain. The ultrasound revealed hemoperitoneum, a uterus with two intramural fibroids and a tender inhomogeneous pelvic mass that seems to connect with the uterus. Computer tomographic (CT) examination raised the suspicion of a degenerated fibroid and hemoperitoneum. Emergency laparotomy was performed: the hemoperitoneum was determined by a degenerated fundal subserosal fibroid. Myomectomy was subsequently carried out. Even though this condition is extremely rare, every clinician has to bear in mind that acute fibroid complications can be a potential cause of acute abdominal pain that requires immediate surgery. The imaging tools, ultrasound and CT are extremely helpful for the diagnosis.

Due to its relative rarity in the second part of the article we have performed a review of the existing literature regarding the acute complications of fibroid torsion.

Keywords: leiomyoma; torsion; hemoperitoneum; fibroid degeneration; acute abdomen

Introduction

Uterine fibroids, also known as myomas or leiomyomas, represent the commonest tumor in women of reproductive age being encountered in approximately 50-60% of women [1-3]. The most accurate term is leiomyoma, the tumor being the result of a monoclonal cell proliferation that originates from the uterine smooth muscle tissue and its connective tissue under the influence of ovarian steroids especially progesterone and local factors [1,2]. Fibroids can remain asymptomatic during the reproductive life being discovered by routine ultrasound (US) or in about 30-40% of cases are diagnosed due to their chronic complications [2].

Fibroid location represents the key factor for the occurrence of a particular complication. Many fibroid classifications are available; the one from FIGO (International Federation of Gynecology and Obstetrics), based on myoma location correlates best with tumor symptoms and treatment [4]. Intracavitary fibroids can determine heavy menstrual bleeding and therefore iron deficiency anemia, but also infertility, while subserosal fibroids can determine pelvic pressure, chronic pelvic pain, urinary or gastrointestinal symptoms or even hydronephrosis [2,5]. The relationship between fibroids and infertility is complex; this type of benign uterine tumors could interfere with fertility through many mechanisms: alteration of endometrial function, increased uterine contractility, impaired endometrial and myometrial blood flow but also alteration of local paracrine factors that can interfere with blastocyst implantation [2].

Extremely rare fibroids can be involved in acute complications such as thromboembolism, acute torsion of subserosal pedunculated leiomyoma, acute urinary retention and renal failure, acute pain caused by red degeneration during pregnancy, acute vaginal or intra-peritoneal

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hemorrhage, mesenteric vein thrombosis and intestinal gangrene [4].

The management of uterine fibroids depends on tumor characteristic: number, size, location of the fibroids but also on patient age and future fertility plans [2]. Surgical therapy is represented by myomectomy or hysterectomy by various approaches: hysteroscopy, laparotomy, laparoscopy or even vaginal surgery. Uterine artery embolization is an alternative to the surgical treatment.

In the following section, we aim to present a rare case of hemoperitoneum due to fibroid degeneration. In the second part of the article we performed a literature analysis regarding imagistic diagnosis and management of fibroids that determined a hemoperitoneum

Case report

A 38-year-old nulliparous woman, with no gynecologic or medical known conditions, presented at the emergency room for severe abdominal pain progressively aggravated in the last two days, despite self-administration of various antalgic and anti-inflammatory drugs. On the day of admission, besides lower abdominal pain, she also presented nausea, dizziness and intense weakness. The patient reported regular menstrual cycles, her last menstrual period being a week ago. She denied intercourse during the last week. The last gynecological examination had been carried out about two years ago and was within normal limits. On physical examination, the vital signs were within normal limits blood pressure 128/84 mmHg, heart rate 76 bpm and temperature of 36.5° C. Abdominal examination showed generalized tenderness and positive Blumberg's sign. Bimanual examination revealed a mobile slightly increased uterus and a tender pelvic mass, about 6 cm in size that could be mobilized together with the uterus.

Hemoglobin concentration was initially 13.6 g/dL and two hours later 12.8 g/dL with leukocytosis ($16.2 \times 10^9/L$). The urine pregnancy test was negative.

US of the pelvis showed uterus into anteversion and antelexion, with two intramural (FIGO 4) fibroids measuring 1.6/1.7 cm and 2.7/3.3 cm and a pelvic mass besides the uterus measuring 6.9/5.8 cm (fig 1a,b). Both ovaries were normal. Moderate free pelvic fluid extending into the pouches of Douglas and Morrison was noted. The repeated US examination after 3 hours revealed the above-mentioned pelvic mass of slightly increased dimensions 8.86/6.22 cm and an increased quantity of free fluid in the peritoneal cavity.

Our initial differential diagnosis was twisted adnexa, ruptured ectopic pregnancy, hemorrhagic corpus luteum or follicular cyst, endometriosis, ruptured ovarian cyst, torsional ovarian fibroma, ruptured pyosalpinx or tubo-ovarian abscess and even appendicitis.

The computer scan (CT) of the abdomen and pelvis showed a 64/88/68 mm mass arising from the fundal wall of the uterus with a vascular pedicle suggesting a fibroid (fig 1c,d).

Due to the detection of hemoperitoneum on paracentesis, corroborated with clinical and imagistic examinations, immediate surgery was performed. Intraoperatively, approximately 400 mL of hemoperitoneum was found originating from the degenerated fundal pediculate fibroid measuring 8 cm, a tumor that has been removed by a myomectomy.

The postoperative recovery was without any incident. Histopathological examination of the mass showed an infarcted and torsion leiomyoma measuring. No increase in mitotic activity has been reported.

The patient was discharged on the fourth day after surgery in the absence of any complication. At 6 weeks postoperative visit the abdominal scar was completely healed, and the ultrasound revealed the presence of the above-mentioned fibroids.

At the scheduled 6-month post-surgery visit, the patient had a viable 6 weeks intrauterine pregnancy.

Fig 1. a) Pelvic ultrasound B mode showing a pelvic tumor related to the uterus and free fluid in the abdominal cavity; b) The pelvic mass was highly suggestive for a uterine fibroid; the lump being in close contact with the uterus (CFM mode); c) CT image suggesting a uterine fibroid, transverse section; d) CT image, sagittal section

Discussions

Spontaneous hemoperitoneum related to fibroids are extremely rare conditions; around 100 cases being reported in the literature [6]. We have performed a search of the literature using the term leiomyoma/fibroids and hemoperitoneum and we found 25 cases reported in the last 10 years (between January 2008 and January 2018) (Table I).

In most of the cases, the condition occurred in women of reproductive age; the mean age of the patients being 35.5 years, ranging from 22 to 62 years. However, Salehi et al [31] reported a case of a 15-year-old adolescent with a 9.5/8.4/10.7 cm heterogeneous, hypoattenuating, symptomatic solid adnexal mass that resulted in being a degenerative uterine leiomyoma. Several other cases in the puerperal period, in perimenopause or even in postmenopause have been reported [18,26].

The majority of complicated leiomyomas were subserosal (92%), among them, 44% being pedunculated. Only one author reported a huge leiomyoma that encompassed the entire uterine corpus. In the latter case, the hemoperitoneum occurred due to the rupture of the dilated superficial vein [24].

The reported leiomyomas complicated with hemoperitoneum varied in size from medium to large dimensions from 4 to 16.3 cm with a median diameter around 11 cm.

The exact etiopathogenetic mechanisms that determine the occurrence of hemoperitoneum in a patient with uterine fibroids are, in some cases, difficult to be identified, the most frequently identified being the rupture of serosal veins or arteries, avulsion, torsion, rupture of a degenerated fibroid.

A precise hemoperitoneum determining cause was reported in several patients: venous congestion, increased abdominal pressure, trauma, rapid growth or degeneration of the fibroid [3,6]. Our patient denied trauma, intense physical activity or intercourse during the last week. She also denies any hormonal treatment or use of oral contraceptives. The only mechanism that we can assume is the rapid growth of a fibroid in a young patient, which overcame its possibilities of vascularization and determined degeneration, later torsion and then rupture of the uterine artery.

Horowitz et al [32] described a case of a 48-year-old woman with a pulsatile bleeding superficial artery, located on the serosal surface of a 14 cm fundal myoma. In this situation, the authors supposed that intraperitoneal hemorrhage could be caused by the underlying pressure exerted by the growing myoma on the walls of a uterine vessel or by the uterine contractions during menstrua-

tion that may distend the blood vessel to the breaking point. Moreover, Lotterman et al [7] described a 28-year-old nulliparous woman that presented hemoperitoneum resulting from surface vein rupture of a large fibroid weighing more than 1300 g. They assumed that the rupture likely occurred after a bowel movement and the increased venous congestion led to vessel rupture in their patient. Schwartz et al [26] concluded that the mechanism responsible for the degeneration and the rupture of a known fibroid in the case of a 53-year-old woman could be related to decreasing levels of estrogen and progesterone in combination with menstrual cycle irregularity.

Rarely, a trauma was depicted in the recent past: Estrade-Huchon et al [11] presented the case of a 46-year-old woman with acute abdominal pain while jogging. Laparoscopy revealed internal bleeding from an avulsed subserosal leiomyoma. In this case abdominal trauma could be responsible for the leiomyoma rupture. By contrast another case of spontaneously avulsion of a uterine leiomyomata in the absence of any trauma was also reported by Pachy et al [10].

Interesting, Swarray-Deen et al [30] described the case of a 43-year-old multiparous woman with massive intra-abdominal hemorrhage due to a 10 cm subserosal fibroid with a ruptured capsule, 2 days after a spontaneous vaginal delivery. It was a leiomyoma with cystic degeneration. The authors concluded that the degenerative process and the contractions during labor could have precipitated the rupture. Moreover, Tan et al [33] reported a similar case but 9 weeks postpartum. In the latter two cases the hormonal changes during postpartum could have determined the degenerative changes and increased the fragility of the tumor.

Several authors [14,21,30,34] reported acute abdomen syndrome after fibroid degenerescence. Three factors have been identified as potential determining for subserosal myoma torsion: the presence of a long and thin pedicle; the size (larger the fibroids less likely to untwist) and the relationship between the myoma and adjacent viscera (the uterine fundus, intestine and pelvic sidewall) [21]. Twisted fibromas causes venous stasis and later on, edema and congestion that will compromise the arterial blood supply. Later hemorrhagic necrosis and gangrene can appear.

Preoperative diagnosis was difficult due to the rareness of the pathology (in our service it was the first case reported in 30 years) and the absence of any medical history (normal gynecologic examination 2 years prior presentation). Imaging techniques (US, CT) helped us to formulate a preoperative diagnosis that was definitely confirmed by the laparotomy which allowed also the treatment (myomectomy).

Table I. Analysis of Pubmed available articles on fibroids and hemoperitoneum (accessed on August 1st 2018)

Authors	Patient age (years)	Type of leiomyoma	Tumor size (cm/kg)	Cause of hemorrhage	Treatment
Lotterman et al (2008) [7]	28	pedunculated, fundal	16/13/10 (1.3 kg)	surface vein bleeding	myomectomy
Ihama et al (2008) [8]	28	subserosal, fundal	10/10/10	rupture of a subserosal vein	an autopsy case
Su et al (2008) [9]	36	subserosal, fundal	15/15/14	dilated vein rupture	myomectomy
Pachy et al (2009) [10]	25	pedunculated anterior	14/10/6	avulsion	myomectomy
Estrade-Huchon (2010) [11]	46	pedunculated, fundal	4 cm	avulsion	hysterectomy
Prior et al (2010) [12]	35	pedunculated, fundal	23/10/17 (3.9 kg)	rupture of serosal veins	myomectomy
Kasum et al (2010) [13]	37	subserosal, fundal, 15 weeks pregnancy	8.5/6.5	ruptured vessel	myomectomy
Foissac et al (2011) [14]	62	pedunculated, fundal	16/23/14	torsion	myomectomy
Toquero et al (2012) [15]	43	subserosal	not mentioned	fibroid laceration	hysterectomy
Chen et al (2013) [16]	22	subserosal	13/13/13	rupture of a superficial UA	laparoscopic myomectomy
Fontarensky et al (two cases) (2013) [17]	47	pedunculated, fundal	10/9/7	no arterial bleeding	embolization of UA, scheduled hysterectomy
Fontarensky et al (two cases) (2013) [17]	44	intramural, anterior	14/14/8.5	no arterial bleeding	embolization of UA, scheduled hysterectomy
Alharbi et al (2013) [18]	55	subserosal fundal	15/12/7	perforation	hysterectomy
Ymeleet al (2013) [19]	46	pedunculated, fundal	22/22/22 (5.5kg)	ruptured varices	hysterectomy
Hicks et al (2014) [20]	33	pedunculated	7.3/6.1/6.5	avulsion	myomectomy
Nigam et al (2014) [21]	42	pedunculated, fundal	9.2/11.7/5.2	torsion	myomectomy
Seet et al (2014) [22]	55	subserosal, fundal	7.3/10/12	ruptured degenerative fibroid	hysterectomy
Peng et al (2015) [23]	39	subserosal, posterior	8/9/9	spontaneous laceration	hysterectomy
Aydin et al (2015) [24]	31	kugel myoma (large leiomyoma which encompassed the whole uterine corpus)	like a 14-16 weeks gestational uterus	rupture of the dilated superficial veins	UA ligation failed, hysterectomy
Gulati et al (2016) [25]	29	pedunculated	14/19/10	ruptured large serosal vessel	myomectomy
Schwartz et al (2017) [26]	53	subserosal, fundal	8.8/7.3/8.3	ruptured degenerated arterial vessel arising from a right UA	hysterectomy
Mizrahi et al (2017) [27]	39	pedunculated, fundal	10/10/10	ruptured degenerated arterial vessel arising from a right UA	myomectomy and ligation of a right UA
Tajima et al (2015) [28]	54	subserosal, posterior	6.5/6/5.5	ruptured arterial aneurysm	myomectomy
Jenayah et al (2017) [29]	37	subserosal, fundal	10/11	ruptured dilated vein	myomectomy
Swarray-Deen et al (2017) [30]	43	subserosal, fundal	10/8	ruptured capsule	myomectomy

UA – uterine arteries

Tsai et al [35] argue that clinical examination together with US is sufficient for a preliminary diagnosis, advocating that no additional advanced radiographic imaging technique is necessary. They justified their option by the fact that emergency surgery must be performed as early as possible to avoid worsening of patient status and avoid consumptive coagulopathy. In our case we considered that the CT scan was helpful and added no significant delay to the patient status, but the use of a CT scan should be considered individualized based also on local resources.

Treatment aims to stop the bleeding and to preserve the uterus, if possible, especially in young women. Myomectomy was reported in 56% of cases (for details see table I); in the rest of the cases, a hysterectomy was performed [11,15,17-19,22-24]. In only two cases, uterine artery embolization was the first line of treatment for patient hemodynamic stabilization followed by a hysterectomy [17].

The immediate intervention is mandatory; the delayed diagnosis and management being associated with increased morbidity and even mortality – one of the reported cases was an autopsy case of a 28 years old woman [8].

Mizrahi et al [27] described the necessity of the ligation of a right uterine artery because of an arterial vessel arising from a right uterine artery with an active bleeding.

Fontarensky et al [17] described the cases of two nulliparous patients with voluminous uterine fibroids complicated by intraperitoneal hemorrhage. The CT showed no arterial blush suggesting active intraperitoneal extravasation and no adnexal mass bleeding. The authors decided to carry out the embolization of the uterine artery, as an emergency therapeutic management, to ensure hemodynamic stabilization before scheduled surgery. Laparoscopic exploration was performed during the following days, but due to the volume of the fibroids the procedure was converted to laparotomy. Moreover, Takeda et al [36] performed gasless laparo-endoscopic single-site myomectomy with in-bag manual extraction for a pedunculated subserosal myoma with a torsion measuring 55/41 mm, without hemoperitoneum.

Conclusions

Hemoperitoneum caused by a uterine fibroid is an extremely rare condition; therefore, it has to be considered as a potential diagnosis after ruling out other more frequent causes of intraperitoneal bleeding in reproductive age women. Imagistic techniques are extremely useful for the diagnosis, US being mandatory. CT can be useful, when available, if the patient is hemodynamically

stable. CT can not only evidence the lesion but also the exclusion of other intra-abdominal and extragenital pathologies. Whenever a fibroid is present in a patient with acute abdominal pain, the rupture of the fibroid feeding vessel should be evaluated. Surgery, regardless of the approach, confirms the diagnosis and allows the immediate bleeding arrest and appropriate treatment (myomectomy, hysterectomy).

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