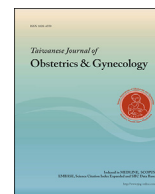




Contents lists available at ScienceDirect

Taiwanese Journal of Obstetrics & Gynecology

journal homepage: www.tjog-online.com

Short Communication

Torsion of pedunculated subserous uterine leiomyoma: A rare complication of a common disease

Yen-Ling Lai ^a, Yu-Li Chen ^{a, b}, Chi-An Chen ^a, Wen-Fang Cheng ^{a, c, d, *}^a Department of Obstetrics and Gynecology, College of Medicine, National Taiwan University, Taipei, Taiwan^b Department of Obstetrics and Gynecology, National Taiwan University Hospital Hsin-Chu Branch, Hsin-Chu City, Taiwan^c Graduate Institute of Oncology, College of Medicine, National Taiwan University, Taipei, Taiwan^d Graduate Institute of Clinical Medicine, College of Medicine, National Taiwan University, Taipei, Taiwan

ARTICLE INFO

Article history:

Accepted 18 July 2017

Keywords:

Abdominal pain

Emergency

Myomectomy

Subserous leiomyoma

Torsion

ABSTRACT

Objective: To evaluate the clinical presentations, diagnosis, management, and outcomes of torsion of the pedunculated subserous uterine leiomyoma.**Materials and methods:** We retrospectively reviewed medical records of patients with subserous uterine leiomyomas undergoing surgeries at National Taiwan University Hospital from January 2001 to December 2015.**Results:** Five cases of torsion of pedunculated subserous uterine leiomyoma were identified. All presented with sudden onset abdominal pain. Two patients received emergent surgeries, the other three cases received scheduled surgeries. The postoperative courses of these five women were uneventful without sequelae.**Conclusions:** Torsion of pedunculated subserous uterine leiomyoma is rare. Accurately diagnosing it prior to surgery is a major challenge. It should be one of the differential diagnosis in patients with uterine leiomyoma presenting with acute abdomen.© 2018 Taiwan Association of Obstetrics & Gynecology. Publishing services by Elsevier B.V. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

Introduction

Uterine leiomyomas (uterine fibroids) are the most common pelvic tumors of reproductive aged women. The prevalence of uterine leiomyoma ranges from 20 to 50% of woman and peaks in their 30s and 40s but declines after the menopause [1]. Considerable evidence indicates that estrogens and progestogens affect the tumor growth [2]. They are classified into intramural, submucous, subserous, cervical, and extrauterine (intraligamentary or intra-ovarian) groups according to the position. Only 50% of women with fibroids exhibit a variety of symptoms, including menorrhagia, dysmenorrhea, pressure symptoms and infertility [1]. It is not common that fibroids could cause acute complications, one of which is acute torsion of pedunculated subserous leiomyoma [3]. Removal of the twisted fibroids usually resolves the problem, but the diagnosis is always intraoperatively confirmed due to limited imaging-based diagnostic characteristics [4]. The purpose of this

study was to review the clinical manifestations, imaging characteristics, management and outcomes of patients diagnosed as having torsion of pedunculated subserous uterine leiomyomas.

Patients and methods

The medical records of patients with subserous uterine leiomyomas undergoing surgeries at National Taiwan University Hospital from January 2001 to December 2015 were retrospectively reviewed. Records were analyzed for demographic details, clinical symptoms, procedural details, pathologic details, and outcomes.

Results

Clinical description

Case 1

A 53-year-old virgin complained of sudden onset diffuse abdominal dull pain for several days. Physical examination revealed diffuse tenderness and rebound tenderness. Laboratory tests showed elevated C-reactive protein level (10.2 mg/dL, normal value < 0.5 mg/dL) with normal white blood cell counts. Transabdominal sonography

* Corresponding author. Department of Obstetrics and Gynecology National Taiwan University Hospital, Taipei, Taiwan. Fax: +886 2 23114965.

E-mail address: wenfangcheng@yahoo.com (W.-F. Cheng).

revealed multiple intramural and one subserous uterine leiomyoma measured 12.0×10.0 cm. The abdomino-pelvic computed tomography (CT) showed multiple uterine fibroids with calcification (Fig. 1A). Exploratory laparotomy showed a necrotic pedunculated leiomyoma with torsion (Fig. 1B and C). Hysterectomy was done smoothly. The pathologic examination showed uterine leiomyoma with focal necrosis (Fig. 1D).

Case 2

This 41-year-old virgin without major medical problem felt right lower quadrant pain for two days. Physical examination revealed diffuse lower abdominal tenderness and rebound tenderness. Transabdominal sonography showed an exophytic uterine leiomyoma (13.0 cm in largest diameter) with degenerated appearance. Laparotomy was performed for acute abdominal pain. A pedunculated subserous fibroid with torsion was noted. Myomectomy was done smoothly. Pathology revealed uterine leiomyoma with focal hemorrhage and necrosis.

Case 3

A 36-year-old nulliparous woman was admitted for acute onset right lower quadrant pain and nausea with stable vital signs. Physical examination revealed diffuse lower abdominal rebound tenderness. The results of blood tests showed normal white blood counts and anemia. Transabdominal sonography revealed one subserous uterine leiomyoma (11.0 cm in largest diameter) and one 5.0×4.0 cm multiloculated cystic right adnexal tumor. Although the total white blood cell counts appeared normal, given the

sonographic appearance and clinical symptoms, tubo-ovarian abscess was highly suspected. Thus, emergent laparotomy was performed. During the operation, a pedunculated uterine leiomyoma with torsion and a right chocolate cyst were found. Myomectomy and right oophorocystectomy were performed smoothly. Ovarian endometriosis and uterine leiomyoma with focal hemorrhage and necrosis were confirmed in the pathology.

Case 4

A 30-year-old woman presented at the emergent department with a complaint of sudden onset lower abdominal pain. Vital signs were stable with normal blood test results. Transvaginal sonography showed one 7.5×7.5 cm subserous leiomyoma and the other 3.6×3.0 cm intramural leiomyoma. Torsion of leiomyoma or degenerative leiomyoma was considered. The symptom subsided after analgesics use. Because the abdominal pain was on and off in the following two weeks, laparoscopic surgery was performed. One 8.0×6.0 cm subserous leiomyoma with torsion and the other 5.0×3.0 cm pedunculated leiomyoma were removed. Leiomyoma with necrosis was confirmed by the pathology.

Case 5

This 36-year-old, nulliparous woman suffered from sudden onset right lower quadrant pain on and off for 2 months. Laboratory tests were within normal limits. Transabdominal sonography showed one 7.0×6.0 cm subserous uterine leiomyoma. Physical and pelvic examination showed mild pelvic tenderness. Analgesics could relieve the pain temporarily, however the similar symptom

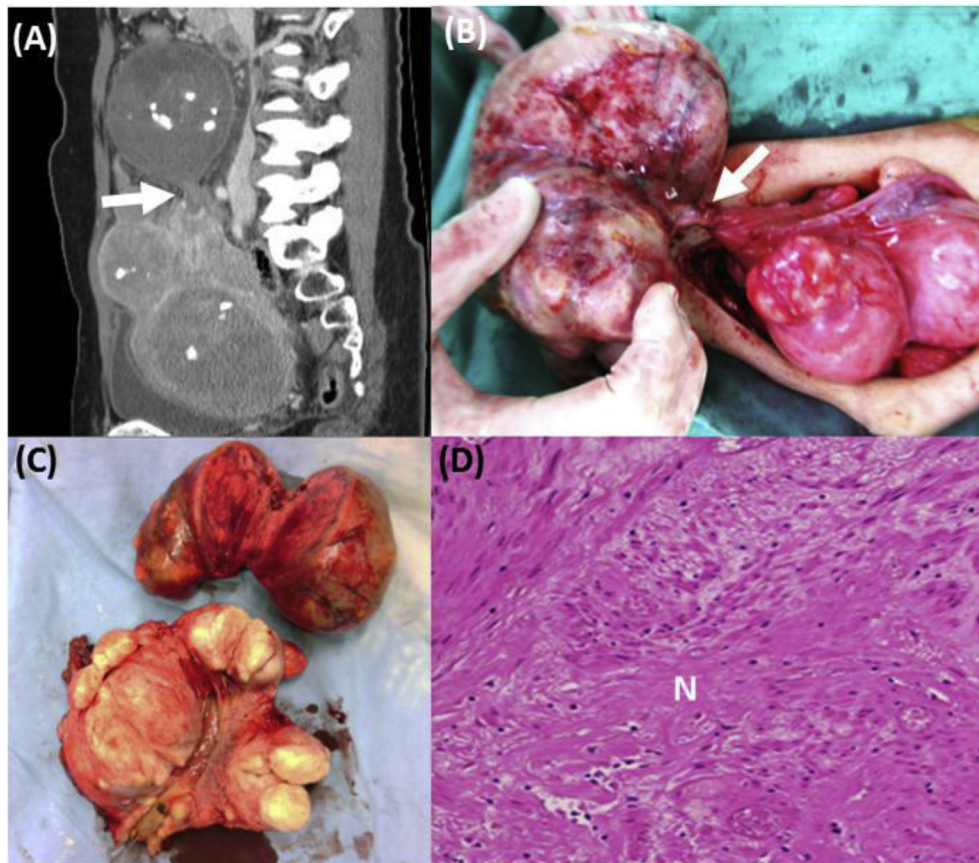


Fig. 1. (A) Sagittal CT scan showed a leiomyoma with a pedicle (arrow) connected to a polymyomatous uterus. (B) A necrotic pedunculated leiomyoma (star) with complete torsion (arrow). (C) Gross view of the cut surface of pedunculated leiomyoma (upper) and uterus (lower). (D) Histologic examination showed leiomyoma with infarct necrosis (N). (hematoxylin and eosin, original magnification $\times 200$).

attacked frequently during the follow-up period. Thus, she received myomectomy and one 7.0 × 6.0 cm pedunculated subserous leiomyoma with torsion was diagnosed intraoperatively. Pathology reported uterine leiomyoma without necrosis and hemorrhage and focal adenomyosis.

Five patients were identified (Table 1.). The median age was 39 years old (range 30–53 years), and all women were nulliparous. All patients presented with acute or frequent abdominal pain. Three cases received myomectomy, one received laparoscopic myomectomy, and the other received total abdominal hysterectomy and bilateral salpingo-oophorectomy. The definite diagnosis were all made intraoperatively. The median size of uterine leiomyoma was 10 cm (7–13 cm). Histologic examinations showed infarct necrosis in four cases and focal adenomyosis in the other one. No post-operative sequela and complication developed during follow-up period in all patients.

Discussion

Torsion of a pedunculated subserous leiomyoma is one of the rare acute complications of uterine leiomyoma [3]. Although patients with uterine leiomyoma may present with severe clinical symptoms, they rarely cause acute complications. However, when they do occur, delayed diagnosis can lead to significant morbidity [5]. Because most cases of torsion of pedunculated subserous leiomyomas were reported as cases in the literature, it is difficult to estimate the incidence of torsion of a pedunculated subserous leiomyoma. From January 2001 to December 2015, 2022 patients surgically diagnosed as having subserous leiomyomas were retrospectively reviewed in the database of our hospital. Only five of the 2022 patients were intraoperatively diagnosed with torsion of a pedunculated subserous leiomyoma. Thus, the incidence of this rare entity is less than 0.25% in patients with subserous uterine leiomyomas undergoing surgical intervention.

The pedunculated subserous leiomyomas are at risk of torsion, especially when the pedicle is thin and long enough to cause excess motility of the leiomyoma and accentuate the torsion. The risk of torsion will increase in the enlarged leiomyoma [6]. However, the cut-off value of the size of leiomyoma for irreversible torsion is uncertain. In our series, three patients (Cases 1, 2, and 3) with subserous leiomyomas larger than 10 cm experienced persistent abdominal pain. Besides, the pain subsided after analgesics used in the other 2 patients (Cases 4 and 5), but it still occurred during follow-up. If the torsion is left untreated, hemorrhagic infarction of the involved leiomyoma may be followed by infection [7].

It is difficult to accurately diagnose torsion of pedunculated leiomyoma before the operation for two main reasons. First, there is no appropriate imaging modality to diagnose a suspected lesion

precisely; second, there are no clinical signs and symptoms that are truly specific for torsion of a pedunculated subserous leiomyoma. Nevertheless, imaging modalities and clinical symptoms may play a role in the detection of torsion of the pedunculated subserous leiomyoma.

Ultrasonography (US) has been employed most frequently in the evaluation of uterine leiomyoma. Twisting of the pedicle on color Doppler may suggest the torsion of pedunculated leiomyoma, but the pedicle of a subserous leiomyoma is not frequently visible because it is often very thin [8]. Roy et al. proposed a diagnosis of acute torsion of subserous leiomyoma when normal ovaries and contrast enhancement of the uterine portion connected to the mass are present on CT [7]. In our five cases, subserous uterine leiomyoma were all diagnosed correctly by ultrasound. Three cases (case 1, 2, and 4) underwent abdomino-pelvic CT, one calcified pedunculated subserous uterine leiomyoma with a pedicle connected to a polypomatous uterus was reported in case 1, another two cases showed subserous uterine leiomyomas.

Magnetic resonance imaging (MRI) is the most accurate imaging technique for detection and localization of uterine leiomyomas. Non-complicated leiomyomas have hypo-intense homogeneous T2 signal and iso-intense T1 signal compared to myometrium [9]. Necrobiotic leiomyomas have heterogeneous hyper-intense T2 signal, and hyper or iso-intense T1 signal depending on hemorrhagic or ischemic mechanism. Sometimes a peripheral halo, hypo-intense on T2 and hyper-intense on T1, may be seen and it corresponds to venous outflow obstruction [10]. While uterine leiomyomas have typical appearance on MRI, pedunculated subserous leiomyomas can simulate an adnexal mass, especially if the stalk is not visualized on the imaging. Reiter et al. provided a practical algorithmic approach to solid adnexal masses and their mimics, with emphasis on the use of anatomic landmarks and imaging characteristics to narrow the differential diagnosis [11].

Although the diagnosis of acute abdomen caused by the torsion of pedunculated subserous leiomyoma is challenging because of its rarity, imaging studies can help to rule out other emergent pathologies, such as acute appendicitis, acute diverticulitis, bowel obstruction, perforated viscus, and adnexal torsion [12].

No obvious abnormal vital signs were detected in our five cases. However, the presence of normal vital signs does not exclude a serious diagnosis. Torsion of pedunculated leiomyoma was confirmed intraoperatively in five cases. No sequelae and complication were found during follow-up after removing the twisted lesion in all five cases. Pain is not a common presenting feature of uterine leiomyoma. However, in some particular conditions, uterine leiomyoma may cause acute pain. The acute sharp pain caused by uterine leiomyoma are usually associated with torsion of the pedunculated leiomyoma, cervical dilatation by submucous

Table 1
Characteristics of five cases of torsion of subserous uterine leiomyoma.

	Age	Parity	Symptom	Time from symptom onset to operation	Imaging findings	Diagnosis	Management	Pathology	Sequelae
Case 1	53	P0	Acute diffuse abdominal pain	2 weeks	US: subserous leiomyoma CT: calcified pedunculated uterine leiomyoma	intraoperative	TAH + BSO	Infarct necrosis	No
Case 2	41	P0	Acute RLQ pain	Hours	US: subserous leiomyoma with degenerative change CT: subserous leiomyoma	intraoperative	Myomectomy	Focal hemorrhage Infarct necrosis	No
Case 3	36	P0	Acute RLQ pain and nausea	Hours	US: subserous leiomyoma	intraoperative	Myomectomy	Focal hemorrhage Infarct necrosis	No
Case 4	30	P0	Acute LLQ pain	2 weeks	US: subserous leiomyoma CT: subserous leiomyoma	intraoperative	LSC myomectomy	Focal hemorrhage Infarct necrosis	No
Case 5	36	P0	Frequent RLQ pain	1 month	US: subserous leiomyoma	intraoperative	Myomectomy	Local adenomyosis	No

TAH: total abdominal hysterectomy, BSO: bilateral salpingo-oophorectomy, LSC: laparoscopic, US: ultrasonography, CT: computed tomography.

leiomyoma protruding through the lower uterine segment, or red degeneration especially associated with pregnancy [13].

In conclusion, the incidence of torsion of pedunculated subserous leiomyoma is rare. Patients with this disease usually present with acute abdomen, stable vital signs, and normal laboratory tests. Despite the imaging advances, the preoperative diagnosis is difficultly made. When a patient presents with acute abdominal pain with a subserous leiomyoma without other possible etiologies, torsion of pedunculated subserous leiomyoma should be kept in the differential diagnosis.

Disclosure

None of the authors has any conflict of interest to declare.

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