

An unusual case of acute abdominal pain: autoamputation of pedunculated subserosal uterine leiomyoma

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Introduction

Uterine leiomyomas are the most common benign neoplasm of the uterus in women of child-bearing age (1). The subserosal type is located beneath the overlying peritoneum (2) and may occasionally cause symptoms, especially in pedunculated lesions, if they twist on their pedicles (3). To our knowledge, torsion and autoamputation of pedunculated subserosal uterine leiomyoma has only been reported twice, once as an incidentally found peritoneal loose body (4) and once as a rare cause of mechanical small bowel obstruction (5) (*Table 1*). Herein, we report a case of an autoamputated pedunculated subserosal uterine leiomyoma in a 47-year-old woman who presented with acute abdominal pain.

Case presentation

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Publication of this article and accompanying images was waived from patient consent according to the Soonchunhyang University Bucheon Hospital institutional review board.

A 47-year-old woman was admitted to the emergency department with aggravating lower abdominal pain.

Physical examination revealed tenderness and rebound tenderness of the abdomen. Laboratory tests showed an elevated white blood cell count of 16,410 cells/ μ L, erythrocyte sedimentation rate of 87 mm/hr, and high sensitivity C-reactive protein of 15.84 mg/dL.

The patient underwent contrast-enhanced abdominal computed tomography (CT). On unenhanced CT, a 7.5×5.3-cm-sized, well-defined, oval shaped, homogenously attenuated mass with focal amorphous calcifications was noted in the right lower abdominal quadrant (Figure 1A). After contrast enhancement, the mass itself showed no contrast enhancement, but thin, rim-like enhancement was noted with surrounding fat infiltrations (Figure 1B). On coronal reformatted images, a pedicle-like structure was noted arising from the mass (Figure 1C). In addition, there were several uterine leiomyomas. Due to the non-enhancing feature, differential diagnosis was focused on various cystic lesions such as complicated mesenteric cyst, mesothelial cyst, or extrapancreatic pseudocyst. Ultrasonography or magnetic resonance imaging was not performed. There was no notable family history, but the patient had a medical history of abdominal CT due to abdominal pain four years prior (Figure 2). At that time, the CT images revealed ascites with pelvic fat infiltrations, suggestive of pelvic inflammatory disease, but the patient was voluntarily discharged. After comparison, we observed a large pedunculated subserosal uterine leiomyoma in the previous

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Authors	Age (years)	Presenting symptoms	Imaging studies	Size (cm)	Surgical procedure
Suganuma <i>et al.</i> (4)	35	Asymptomatic, incidentally detected mass in pelvic ultrasound	Ultrasound, magnetic resonance imaging	6.0×7.0	Exploratory laparoscopy
Rubinkiewicz <i>et al.</i> (5)	70	Bowel obstruction with abdominal distension, muscle guarding and rebound tenderness	Ultrasound	20.0×10.0	Exploratory laparotomy
Present case	47	Aggravating abdominal pain	Computed tomography	7.5×5.3	Exploratory laparotomy

\mathbf{I}	Table 1 Literature	review of autoampu	utated pedunculate	d subserosal u	iterine leiomvo	oma
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Figure 1 CT images of autoamputated pedunculated subserosal uterine leiomyoma. (A) Unenhanced axial CT image shows a 7.5×5.3-cmsized, well-defined, oval shaped, homogeneously attenuated mass (arrow) with a focal area of amorphous calcifications (arrowhead). After contrast enhancement, (B) on axial CT image of the portal phase the mass shows thin and even-thickness rim-like enhancement (arrowhead) and no contrast enhancement in the mass itself. (C) On coronal reformatted CT image of the portal phase, there was a pedicle-like structure (arrowhead) arising from the mass (arrow). Also, in the uterus, there were several heterogeneously enhancing masses (black asterisk), which were diagnosed as uterine leiomyomas. CT, computed tomography.

CT that was not currently visible. The patient had no history of myomectomy during the prior four years. Thus, autoamputation of pedunculated uterine leiomyoma was suspected, and due to the large size, exploratory laparotomy was performed. A huge mass with adhesion to the omentum was noted with no ongoing bleeding. The mass was easily removed during the one-hour surgical procedure.

The specimen showed a typical whirling pattern of leiomyoma and homogeneous dark red appearance

compatible with red degeneration on the cut surface (*Figure 3*). Under the microscope, the center of the mass was hyalinized with a few viable spindle cells in the periphery. The tumor was positive for desmin immunohistochemical stain, supporting the diagnosis of leiomyoma.

Discussion

To our knowledge, this is the first reported case of an



Figure 2 Contrast-enhanced CT image taken four years prior shows heterogeneously enhancing uterine leiomyomas in the intramural (black asterisk) and subserosal (white arrow) areas of the uterus. In the large subserosal uterine leiomyoma, a pedicle (black arrowhead) is seen connecting the mass to the uterine body. CT, computed tomography.

autoamputated subserosal uterine leiomyoma, presenting as a cause of acute abdominal pain. Uterine leiomyomas may sometimes be symptomatic, and abdominal pain may occur in cases of degeneration, torsion, or vaginal bleeding (3). Pedunculated subserosal uterine leiomyomas are morphologically attached to the uterus by a stalk or pedicle, making them susceptible to torsion. Abdominal CT and ultrasonography are the main imaging modalities for evaluation, but the diagnosis may be challenging when the leiomyoma has twisted and completely detached from the uterus.

Previous studies have reported CT findings of subserosal uterine leiomyomas with acute torsion (6,7). Ohgiya *et al.* (6) and Roy *et al.* (7) reported that poor or no contrast enhancement of the leiomyoma, regular thin rimenhancement, fan-shaped poor contrast enhancement in the uterus adjacent to the leiomyoma (dark fan sign) or a



Figure 3 Pathology of autoamputated pedunculated subserosal uterine leiomyoma. Macroscopic (A) and microscopic findings with hematoxylin and eosin staining: (B) 100×, (C) 200× magnification; and immunohistochemical staining for desmin: (D) 100× magnification. (A) Gross findings showed that the tumor is a well-defined ovoid mass surrounded by omental fat tissue. The cut surface of the mass shows a typical whirling pattern of a leiomyoma, with homogeneously dark red color due to red degeneration. (B) Most of the mass showed hyaline degeneration under the microscope, and spindle cells were observed in some areas of the periphery (C). (D) The mass was generally positive on desmin staining.

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localized shaggy zone with intense enhancement between the leiomyoma and the uterus were valuable features for identifying acute torsion of the subserosal uterine leiomyoma.

When the pedicle of a subserosal uterine leiomyoma is twisted, venous congestion and edema occur causing compression of the arterial blood flow, and no contrast enhancement of the mass itself on CT. In time, hemorrhagic infarction and necrosis will occur, leading to infection and peritonitis, if left untreated. Ohgiva et al. (6) and Roy et al. (7) had different explanations for the rim-enhancement of the uterine leiomyoma with acute torsion. Roy et al. (7) suggested that the rim corresponds to the obstructed peripheral veins of dilated lymphatic vessels, while Ohgiya et al. (6) suggested it may be the collateral capsular perfusion. In our case, a similar rim-enhancement was observed, but neither of these hypotheses was appropriate because the leiomyoma was intra-abdominalfree. Considering the surrounding fat infiltrations on CT and adhesion of the mass with omental fat on the surgical specimen, we speculate that the rim-enhancement was due to peritonitis caused by the necrotized autoamputated uterine leiomvoma.

Lastly, direct or indirect imaging features of the twisted pedicle is a valuable clue that suggests acute torsion of the pedunculated subserosal uterine leiomyoma. Ohgiya et al. (6) speculated that the dark fan sign reflects decreased perfusion of the uterine muscles due to compression by a rigid, twisted pedicle. Roy et al. (7) surgically confirmed that the shaggy enhancement between the uterus and the avascular mass was the twisted pedicle itself. In our case, a pedicle was seen arising from the avascular mass. suggesting that the mass was originally attached to another organ. There have been reports of large peritoneal loose bodies originating from sequestered ovaries (5), tumors of ovarian origin (8,9), or pedunculated subserosal uterine leiomyomas (4). In our case, a large pedunculated subserosal uterine leiomyoma in the previous CT was not currently visualized, supporting our judgment that the mass was most likely an autoamputated subserosal uterine mvoma.

Our report highlights a rare case of autoamputated pedunculated subserosal uterine leiomyoma presenting as a cause of acute abdominal pain. Such imaging diagnosis may be challenging due to complete detachment of the leiomyoma from the uterus. However, a careful comparison with previous imaging studies and imaging features such as a pedicle attached to the main avascular mass may help accurately diagnose such a rare case.

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Footnote

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://qims. amegroups.com/article/view/10.21037/qims-23-1538/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Publication of this article and accompanying images was waived from patient consent according to the Soonchunhyang University Bucheon Hospital institutional review board.

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