Conservative Management of Uterine Artery Pseudoaneurysm Rupture with Rectal Fistula after Laparoscopic Myomectomy and Review of the Literature

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Abstract
Late uterine hemorrhage caused by uterine artery pseudoaneurysm is a rare but life-threatening complication after uterine surgery. The association of uterine pseudoaneurysm rupture causing rectal fistula has not been reported in literature as well as its management. Herein we report a case of massive late uterine bleeding and rectal bleeding after laparoscopic myomectomy.

A 43 year old Caucasian nulliparous woman underwent laparoscopic excision of 8 cm posterior intramural myoma. One month later she developed acute abdominal pain and proctorrhagia. A Computer Tomography scan (CT) demonstrated a 10 cm pelvic hematoma. The transvaginal ultrasound (TVU) showed 1 cm aneurism of the uterine artery. Transarterial angiography confirmed the ruptured uterine artery pseudoaneurysm that was successfully embolized. Repeated colonoscopy showed a rectal fistula. Total parenteral nutrition (TPN) and antibiotic prophylaxis were started. During the following month the intensity and frequency of proctorrhagia gradually declined. After four week of TPN colonoscopy confirmed complete fistula healing. Oral feeding was gradually reintroduced. Two months later an ultrasound scan showed a complete resolution of the pelvic hematoma. To the best of our knowledge this the first described association of two rare complications: a pseudoaneurysm rupture and the subsequent rectal laceration successfully treated with embolization and conservative management.

Keywords
Uterine artery pseudoaneurysm, Rectal bleeding, Rectal fistula, Myomectomy

Introduction
Pseudoaneurysm of uterine artery (UAP) is a rare complication after gynecological, obstetric and pelvic surgery. Pseudoaneurysm is characterized by the absence of the three layers in its boundaries and by the presence of a peripheral thrombus [1] and this is the main difference with true aneurysm. The incidence of pseudoaneurysm formation is not well known because it can be silent, the delay of symptoms if rupture occurs [1] and the diagnosis is not easy. This event can be life-threatening if rupture happens. Isono [2] found that the interval between surgery and pseudoaneurysm rupture can vary between 3 to 60 days.

The main symptoms of rupture are methorrhagia, abdominal pain and anemia. Diagnosis can be made with Ultrasonography (US), Color Doppler Ultrasonography (CDUS), Computer Tomography (CT), Magnetic Resonance Imaging (MRI) and angiography. The gold standard for treatment is transfemoral angiography with concomitant embolisation. We report a case of pseudoaneurysm rupture associated with bleeding from the rectum and conservative management along with a review of literature. A PubMed search of English literature was performed using: “uterine artery pseudoaneurysm” and “uterine artery pseudoaneurysm and rectal bleeding”. We found 106 articles, 105 for the first search and only one for the second. The latter refers to a case of locally advanced cervical [3]. In our knowledge our case should be the first of a ruptured UAP, after laparoscopic myomectomy, associated with proctorrhagia.

Case Report
A 43 years old nulliparous woman with sudden abdominal pain associated with rectal bleeding came to our attention in April 2013. A gasless laparoscopic myomectomy had been performed one month before (in another hospital) for the removal of an 8 cm intramural leiomyoma, located on the posterior wall, causing severe methorrhagia. Although proctorrhagia was not massive her hemoglobin concentration at admission was 10,4g/l, dropping to 8,4g/l after 3 hours. Leucoctysis was also present. She immediately underwent a CT scan showing the presence of a dyshomogenous mass behind the uterus of 10x7x8cm with a central hypodense part.
pseudoaneurysm was confirmed and embolisation was performed.

catheters introduced in the right femoral artery. During the exam the
surgeon we decided to perform transfemoral angiography with 4 Fr
patient remained hemodynamic stable. After counseling with general
hyperecogenic mass of 8 cm. The anechoic part was pulsing. The
the posterior wall of uterus discontinued with an adjacent hypo-
area was found presenting no clear cleavage plane from the bowel.
( hematic). In the iliac fossa and above the rectum an hemorrhagic
The sigmoid tract appeared swalled and hyperhemic. Free fluid
CT: Computerized Tomography; CRPL: C-reactive protein; MRI: Magnetic Resonance Imaging; TVUS: Transvaginal Ultrasound; CDUS: Color Doppler Ultrasound

Table 1: Pseudoaneurysms of uterine artery after myomectomy reported in the literature (up to date December 2014)

<table>
<thead>
<tr>
<th>Study</th>
<th>Surgery</th>
<th>Surgical indications</th>
<th>Age at surgery, yrs</th>
<th>Gravity</th>
<th>Signs and symptoms caused by uterine artery pseudoaneurysm</th>
<th>Interval between surgery and symptom onset</th>
<th>Diagnostic procedures</th>
<th>Initial diagnosis</th>
<th>State of pseudoaneurysm at diagnosis</th>
<th>Size, mm</th>
<th>Location of pseudoaneurysm</th>
<th>Treatment procedure</th>
</tr>
</thead>
<tbody>
<tr>
<td>Wan et al [7]</td>
<td>Abdominal myomectomy</td>
<td>Menorrhagia</td>
<td>38</td>
<td>N/A</td>
<td>hemorrhage</td>
<td>22d</td>
<td>CT scan</td>
<td>N/A</td>
<td>N/A</td>
<td>Transarterial embolization</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Ito et al [9]</td>
<td>Hysteroscopic myomectomy</td>
<td>Menorrhagia</td>
<td>39</td>
<td>G1 P1</td>
<td>Massive uterine bleeding</td>
<td>3 d</td>
<td>CT scan</td>
<td>Ruptured pseudoaneurysm</td>
<td>Ruptured</td>
<td>10</td>
<td>Right uterine artery</td>
<td>Transarterial embolization</td>
</tr>
<tr>
<td>Higgin et al [10]</td>
<td>Abdominal myomectomy</td>
<td>Intramural myoma</td>
<td>40</td>
<td>G1 P1</td>
<td>Sudden and abundant metrorrhagia</td>
<td>40 d</td>
<td>TVUS,CDUS, pelvic arteriography</td>
<td>Ruptured pseudoaneurysm</td>
<td>Ruptured</td>
<td>30</td>
<td>Left uterine artery</td>
<td>Transarterial embolization</td>
</tr>
<tr>
<td>Takada et al [9]</td>
<td>Laparoscopic-assisted myomectomy</td>
<td>Intramural myoma</td>
<td>32</td>
<td>G0 P0</td>
<td>Sudden massive uterine hemorrhage at menstruation</td>
<td>79 d</td>
<td>TVUS, pelvic arteriography</td>
<td>Massive uterine hemorrhage of unknown origin</td>
<td>Ruptured</td>
<td>29</td>
<td>Peripheral branch of left uterine artery</td>
<td>Transarterial embolization</td>
</tr>
<tr>
<td>Takada et al [12]</td>
<td>Laparoscopic-assisted myomectomy</td>
<td>Intramural myoma</td>
<td>32</td>
<td>G1 P0</td>
<td>Fever elevated CRP</td>
<td>3 d</td>
<td>Routine post-operative TVUS, CDUS, CT</td>
<td>Pseudoaneurysm</td>
<td>Unruptured</td>
<td>27</td>
<td>Right uterine artery</td>
<td>Transarterial embolization</td>
</tr>
<tr>
<td>Asai et al [13]</td>
<td>Laparoscopic-assisted myomectomy</td>
<td>Infertility</td>
<td>36</td>
<td>G0</td>
<td>Asymptomatic, Metrorrhagia</td>
<td>120 d, 4 d</td>
<td>Routine post-operative TVUS, CDUS, MRT, CT</td>
<td>Pseudoaneurysm</td>
<td>Unruptured</td>
<td>53</td>
<td>Left uterine artery</td>
<td>Transarterial embolization</td>
</tr>
<tr>
<td>Current report</td>
<td>Laparoscopic gasless myomectomy</td>
<td>Intramural myoma</td>
<td>43</td>
<td>G0</td>
<td>Abdominal pain, rectal bleeding, anemia</td>
<td>30 d</td>
<td>TVUS, CT</td>
<td>Pelvic arteriography</td>
<td>Pseudoaneurysm</td>
<td>Ruptured</td>
<td>-</td>
<td>Left uterine artery</td>
</tr>
</tbody>
</table>

CT: Computerized Tomography; CRPL: C-reactive protein; MRI: Magnetic Resonance Imaging; TVUS: Transvaginal Ultrasound; CDUS: Color Doppler Ultrasound

Angiographic control performed during procedure showed an important active bleeding from the pseudoaneurysm and so cyanoacrylate 1 ml was injected before metallic spirals to achieve complete embolization (Figure 1,2).

Long term conservative management was decided along with the patient also in view of lack of interest for future pregnancy. Otherwise due to possible sequelae on fertility arising from severe pelvic infection such a decision should be taken only after a thorough discussion with the patient. Colonoscopy revealed the presence of rectal fistula explaining the observed procthorragia. Parenteral antibiotic therapy with Piperacillin and Tazobactam (2g+0,25g) and nutrition was administered daily. Abdominal pain decreased and

Figure 1: Angiographic finding of bleeding left UAP

Figure 2: Left UAP embolisation
stopped in few days while procthorragia disappeared in one week. Hemoglobin concentration raised and leucocitosis normalized in two weeks. Colonoscopy, repeated one month later, revealed the complete healing of the rectal fistula allowing a return to enteral nutrition. Two months after hospital discharge a normal pelvic finding with no visible hematoma was observed at follow-up TVU scan.

Discussion

This is the first described case of UAP rupture associated with rectal bleeding. The treatment of the pseudoaneurysm with transfemoral angiography and embolisation is the gold standard either to confirm the diagnosis or to treat the hemorrhage. In our case the expectant management avoided major surgery such as hysterectomy, bowel resection and protective colostomy. Few cases of pseudoaneurysm are reported in the literature. This is due to the rare frequency and also the difficult diagnosis.

Table 1 shows the summary of the 9 reported cases of postgynecologic surgical uterine artery pseudoaneurysms (including current case) found on PubMed search. Two cases occurred after hysterectomy 3 cases occurred after laparotomic myomectomy and the other 2 after laparoscopic myomectomy for intramural myoma. It is interesting to know that, on TVU scan, pseudoaneurysms have characteristic sonographic appearance consisting of pulsating anechoic or hypoechoic, well-defined cystic structure. This can be found with or without an associated pelvic hematoma or free fluid. Although color Doppler ultrasonography might help in establishing the diagnosis (by revealing blood flow within cystic structure after initial identification on gray-scale ultrasonography) one must remember that sensitivity of color Doppler is variable depending on the site of pseudoaneurysm [4]. Emergency angiography remains the gold standard for definitive diagnosis. Endovascular embolization is now the preferred method of management pelvic pseudoaneurysms [5,6] as shown in the reported 9 cases (Table 1). This technique has significant advantages over traditional surgery.

References