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Case Report

Uterine artery pseudoaneurysm: A rare cause of postmyomectomy bleeding: Case report and literature review

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ABSTRACT

Uterine artery pseudoaneurysm (UAP) is an acquired abnormal blood vessel like space formed from a leak of a uterine artery branch and accumulation of thrombus. Most UAPs occur inside the uterus. They occur rarely and can develop after various gynaecologic and obstetric procedures. Delay in its diagnosis can result in life threatening haemorrhage.

Here we present a case of a 48-year-old woman who presented with heavy vaginal bleeding one month after removal of multiple uterine myomas. Ultrasound and color doppler showed a uterine artery pseudoaneurysm. Emergency subtotal hysterectomy had to be performed due to catastrophic bleeding. She was found to have a ruptured uterine artery pseudoaneurysm at one site of myoma removal and an intact uterine artery pseudoaneurysm at another site of myoma removal. Literature review was done and this case was compared with other cases of delayed post myomectomy haemorrhage and uterine pseudoaneurysms to find out the presentation, differential diagnosis and management of delayed post myomectomy haemorrhage and uterine artery pseudoaneurysms.

Keywords: Myomectomy complications, Uterine artery pseudoaneurysm, Post myomectomy haemorrhage, Uterine vascular lesions, Uterine vascular malformations

INTRODUCTION

A pseudoaneurysm is defined as dilatation of an artery with partial or full disruption of the vessel wall (Karmous N, 2016). It can be caused by a traumatic injury to the vessel wall with subsequent formation of periarterial haematoma that liquefies forming a periarterial blood cyst. It communicates through a narrow neck with the arterial lumen (Youssef AT, 2018). The differential diagnosis of vaginal bleeding after a myomectomy include abnormal uterine bleed, a bleeding haematoma and uterine vascular lesions like uterine artery pseudoaneurysm (UAP), arteriovenous fistulas (AVF) or arteriovenous malformations (AVM) (Jennings L, 2019). Reintervention rates after abdominal myomectomy have been seen from 14 to 25% (Davis MR, 2018). Delayed diagnosis of iatrogenic vascular abnormalities like UAP, AVF and AVM may worsen bleeding if treatment is pursued for alternative diagnosis. Being aware of such vascular conditions and how to diagnose them will allow early proper treatment and more favorable outcome (Jennings L, 2019).

Transvaginal ultrasound aided with colour doppler capability is an accurate tool for assessment (Youssef AT, 2018). In this report, we present a case of UAP formation one month after removal of myomas at two sites in the uterus. One was found intact and the other had ruptured and caused catastrophic bleeding.

CASE REPORT

A 48-year-old lady with a previous one caesarean section and one normal delivery had abdominal myomectomy and mirena insertion for multiple uterine fibroids at our hospital. A large subserosal myoma of 11 x 13 cm and multiple intramural fibroids were removed one of them of 3.3 cm in size from left posterolateral area which was displacing endometrial cavity anteriorly. Intraoperative and postoperative period was uneventful and she was discharged on 2nd postoperative day in a healthy condition. Her postoperative visits were also uneventful. Exactly one month after her operation she presented to emergency department with complaint of on and off episodes of

heavy vaginal bleeding for one week with vaginal spotting in between, two episodes of heavy vaginal bleeding during the day and an episode of heavy bleeding with passage of clots and fainting at midnight for which she came to the emergency.

On examination she had stable vitals, abdominal examination was normal and speculum examination showed a small clot and the mirena thread protruding from the cervical os. Hb was 9.6 g/dl, Ultrasound showed a 3.5 X 4 cm sac like structure filled with actively flowing blood in the lower uterine area with suspicion of uterine pseudoaneurysm (Figure 1-3). Cervix was closed with intrauterine device seen inside the cervical canal, there was mild free fluid in the pelvis with both ovaries normal.

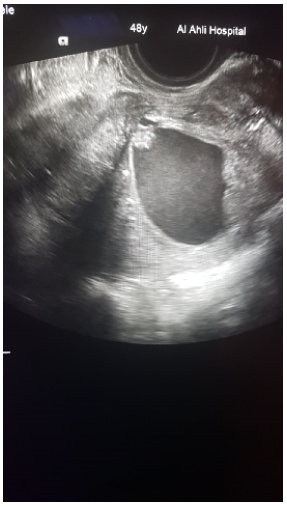

Figure 1. Gray scale transvaginal ultrasound image of uterus showing an anechoic mass

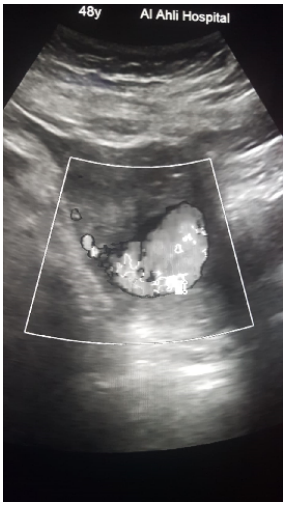

Figure 2. Color doppler image of vascular lesion showing turbulent blood flow with varying colors

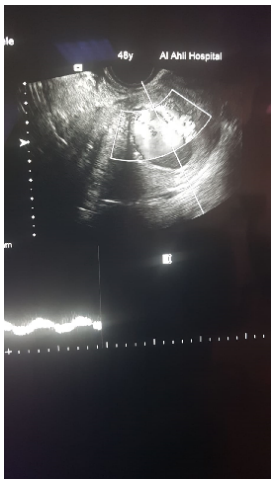

Figure 3. Power doppler image showing turbulent blood flow

She was admitted and given inj tranexamic acid 1 g i/v, normal saline infusion, crossmatched packed RBCs were requested and inj ceftriaxone 1 g i/v bid and inj Metronidazole 500 mg i/v TID were started. On removing mirena there was severe bleeding. Hysteroscopy was unsuccessful due to heavy bleeding. Curettage revealed a clot and some macerated tissue. Still there was continuous severe bleeding.

Subtotal hysterectomy with bilateral salpingectomy was done. All previous myomectomy scars in the uterus seemed to be normal. No cause for bleeding could be found on gross examination of uterus. She received 2 units of PRBC and 2 units of fresh frozen plasma due to severe bleeding. Postoperative Hb was 7.8 g/dl. One further unit of PRBC was transfused.

The histopathological report revealed secretory endometrium. A cyst was seen of about 1.8 cm in right cornual area partially lined by stretched endometrium that gave positive CD31 immunostain indicating its vascular nature: This cyst was adjacent to the site of removal of the big subserous myoma (figure 4).

Figure 4. Photograph showing the uterine pseudoaneurysm in left uterine cornua

On the left side of the uterus there was a cavity in the myometrium adjacent to the endometrium and showing a tract connecting to endometrial cavity surrounded by glandular tissue, blood, fibrin and chronic inflammatory cells with many foreign body giant cells. This cavity corresponded in size and area to the cystic lesion with actively flowing blood seen on ultrasound and it was in the area of previous myoma removal and was surrounded with sutures (Figure 5).

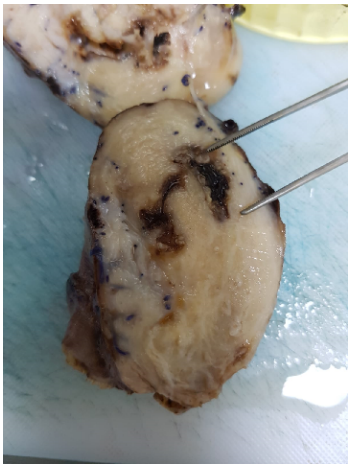

Figure 5. Photograph showing ruptured uterine pseudoaneurysm in the left side of the uterus connecting to endometrial cavity through a tract

She was discharged on 3rd postoperative day in a good condition with a healthy wound and a Hb of 8.5 g/dl. Her postoperative follow up was uneventful.

DISCUSSION

Increasing the number and size of the fibroids increases the risk of haemorrhage after a myomectomy (Tanos V, 2018). The information on differential diagnosis of delayed vaginal bleeding after myomectomy is lacking in literature. It includes causes of abnormal uterine bleed like polyps, adenomyosis, leiomyoma, hyperplasia, malignancy and coagulopathies and vascular lesions like arteriovenous malformations, arteriovenous fistulas and uterine artery pseudoaneurysm (Hapamgaman DK, 2016).

Delayed diagnosis may worsen bleeding in the setting of a ruptured UAP or AVM if treatment is pursued for alternative diagnosis. Being aware of UAP and AVM will allow early proper treatment and more favorable patient outcome (Jennings L, 2019).

A pseudoaneurysm is an extraluminal collection of blood with turbulent flow that communicates with the parent vessel through a defect in the arterial wall. The absence of a 3 layered arterial wall lining the pseudoaneurysm differentiates it from a true aneurysm, which is less common. UAP might appear in the form of a small vascular malformation at a very early stage (Higon MA, 2007, Langer JE, 1999).

UAPs can be asymptomatic or cause life threatening vaginal or intraabdominal bleeding when ruptured. Thus, these pseudoaneurysms should be considered in differential diagnosis of any unexplainable vaginal bleeding especially after a recent obstetric or traumatic procedure (Ros C, 2017).

The most frequent cause of UAP was caesarean section, which accounted for 47.4% of all cases. Other causes include dilatation and curettage and non-traumatic vaginal delivery (Isono N, 2010, Wang LC, 2016). Mean interval between the incident and the symptoms was approximately two weeks regardless of the cause (Isono N, 2010).

Acquired AVMs are characterized by multiple unions of varying sizes between arteries and veins in the same vicinity. Affected patients commonly present with menorrhagia or menometrorrhagia after a miscarriage, uterine surgery or curettage. Symptoms can appear very slowly or suddenly. Bleeding is often intermittent and torrential. This is secondary to high vascular flow across the arterial and venous systems (Yoon DJ, 2016). In a study on evaluation of post myomectomy scar by Darwish AM et al. Hematoma in the myomectomy bed was observed postoperatively in 24%, 17% and 7% patents on day 2,7 and 1 month respectively (Darwish AM, 2005).

Diagnosis of pseudoaneurysm is made with doppler ultrasound, which shows swirling arterial flow in different directions and velocities with varying colors according to the degree of turbulence within the pseudoaneurysm (Hennerici M, 1999). The ultrasonographic characteristics of AVM are nonspecific and include the presence of hypoechogenic tubular structures within the myometrium by gray scale ultrasound imaging. The identification of uterine high velocity blood flow with low impedance by doppler ultrasound is highly suggestive for a uterine AVM. Adenomyosis in the setting of menorrhagia also can have a similar ultrasound finding such as hypervascularity and turbulent flow (Timmerman D, 2003).

Our patient presented to emergency department with intermittent heavy vaginal bleeding for a week with spotting in between and severe bleeding episode on presentation. Ultrasound showed a 3.5 x 4 cm sac like structure filled with actively flowing blood in the lower segment. Cervix was closed with displaced mirena in the cervical canal. The differential diagnosis included a uterine vascular lesion like a uterine artery pseudoaneurysm.

The presentation was similar to other reports. In one case report by Higon et al a 40 year old woman

presented at 40 days post myomectomy with sudden severe metrorrhagia with similar ultrasound findings (Higon MA, 2007). In another report a woman presented 19 days after dilatation and evacuation for elective termination of pregnancy with heavy bleeding due to a UAP (Jennings L, 2019). Another patient presented 1 month after a caesarean section with similar complaints (Boi L, 2017).

Rupture of a vascular abnormality including congenital AVMs, True aneurysms and pseudoaneurysms can lead to a life-threatening situation. Curettage of an unknown vascular abnormality can lead to massive haemorrhage (Wang LC, 2016). Traditional surgical management of UAPs includes revision with packing, bilateral internal iliac or uterine artery ligation and when other treatments fail, hysterectomy. Transcatheter arterial embolization has recently emerged as a safe and highly effective alternative treatment (Boi L, 2017). Surgery with vessel and uterine repair or hysterectomy has been reported as the definitive treatment for pelvic bleeding related to myomas and post myomectomy complications with massive haemorrhage (Lotterman S, 2008).

Our patient had an episode of heavy vaginal bleeding and was taken to operation theatre for removal of displaced mirena and hysteroscopy. On removing the displaced mirena there was severe bleeding. Hysteroscopy did not show anything due to heavy bleeding. Curettage was done, a clot and some macerated tissue was removed. There was continuous severe bleeding after curettage. Subtotal hysterectomy with bilateral salpingectomy was done. The histopathology of the uterus showed secretory endometrium. A uterine artery pseudoaneurysm in the area of the right cornua filled with blood measuring 1.8 cm possibly resulted from previous myomectomy, as it was very near to multiple foreign body giant cell granulomas at the site of myoma removal. The area of myometrial haemorrhage on the left side of the uterus showed a cavity in the myometrium adjacent to the endometrium with a tract connecting to endometrial cavity surrounded by glandular tissue, blood, fibrin and chronic inflammatory cells with many foreign body giant cells. This cavity corresponded in size and area to the cystic lesion with actively flowing blood seen on ultrasound and it was in the area of previous myoma removal and was surrounded with sutures. It was presumed that this vascular lesion is most probably a uterine pseudoaneurysm in view of its clinical presentation of intermittent heavy bleeding, ultrasound and doppler findings of a cystic lesion with actively flowing blood and histopathological finding of a cavity in the myometrium adjacent to many blood vessels in the area of previous myoma removal.

CONCLUSION

In conclusion this case illustrated a rare but serious complication of gynecologic surgery. It shows two uterine vascular lesions one of them a histologically confirmed uterine artery pseudoaneurysm and the other a ruptured vascular lesion most probably also another uterine pseudoaneurysm. These lesions presented with heavy bleeding one month after a myomectomy with mirena insertion.

Early diagnosis could have prevented the hysterectomy and the UAP could have been managed with selective arterial embolization. It shows the importance of post myomectomy follow up for any rare vascular complications like UAPs which can lead to catastrophic bleeding.

CONFLICTS OF INTEREST

The authors declare no conflict of interest regarding the publication of this case report. Consent was obtained from the patient regarding the publication of her medical history, investigations and management.

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