CASE REPORT

Uterine artery pseudoaneurysm following laparoscopic hysterectomy. An unusual cause of delayed heavy vaginal bleeding

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Introduction

Uterine artery pseudoaneurysms are rare acquired vascular malformations that can develop after vascular injury, inflammation or following pelvic surgery such as caesarean section, uterine curettage or hysterectomy [1]. They can undergo spontaneous rupture and present with unexpected intra-abdominal or vaginal bleeding, metrorrhagia or an infected pelvic collection.

Case presentation

This is the case of a 36-year-old woman, para 3, who underwent total laparoscopic hysterectomy and left salpingo-oophorectomy for pelvic pain. Her symptoms developed 12 months after a NovaSure endometrial ablation for menorrhagia. Pre-operatively, she had a trial of 3 months GnRH analogues with complete resolution of pain. Intraoperative findings included a normal-sized uterus, adhesions of the left ovary to the pelvic side wall and extensive adhesions of the bladder to the cervix as a result of three previous caesarean sections. The procedure was uncomplicated with blood loss of 100 ml. A vessel sealing device was used for haemostasis (Gyrus PK, Olympus) and the vagina vault was closed with V-loc 2–0 barbed suture (Covidien).

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A. Page Department of Interventional Radiology, Royal Hampshire County Hospital, Winchester, UK Post-operative course was uneventful and she was discharged home on day 1.

She was readmitted 22 days post-operatively with sudden onset of heavy vaginal bleeding with big clots. On admission, there was no abdominal tenderness; she was apyrexial and haemodynamically stable. The admission haemoglobin was 13 g/dl, and inflammatory markers were normal. On speculum examination, there was arterial bleeding from the left angle of the vaginal vault, and further clots were removed from the vagina. The estimated blood loss was 700 ml. Our standard protocol for readmissions usually involves an ultrasound scan and/or CT scan depending on the clinical indication. However, in this case, due to the continuation of brisk bleeding, it was decided to transfer her to theatre before any imaging was performed. In the operative theatre, four figure of eight haemostatic sutures were placed in the vaginal vault achieving complete haemostasis. Due to the complete arrest of vaginal bleeding, we did not consider that a laparoscopy was necessary. A vaginal pack with antiseptic proflavine was inserted in the vagina for 24 h. She was transfused with two units of red blood cells, and the post-transfusion haemoglobin was 9.5 g/dl. Her immediate post-operative course was stable, but the following day after removal of the pack, bleeding resumed with a further 200 ml of fresh vaginal loss.

In order to avoid further surgery, she underwent an emergency pelvic angiogram with a view of embolisation of the pelvic vessels. Initial flush angiography did not clearly demonstrated vascular abnormality. Selective angiography of the anterior division of the left internal iliac artery and superselective angiography of the uterine artery demonstrated a pseudoaneurysm arising from the distal residual left uterine artery (Fig. 1).

The left uterine artery was cannulated using a Terumo Progreat micro catheter (Terumo Corp. Tokyo, Japan) via a 5 French Sim2 catheter as a guide (Boston Scientific,



Fig. 1 Superselective left uterine artery angiogram demonstrating a pseudoaneurysm of the distal artery (thin arrow) and post-procedural angiogram showing complete vessel occlusion and reflux into other internal iliac artery branches (thick arrow)

Natick, USA). Three ampoules of $355-500 \ \mu m$ PVA (Contour, Boston Scientific) and six $4 \times 4 \times 20$ mm micro coils (Boston Scientific) were used for the embolisation. This achieved complete occlusion of the vessel (Fig. 1). Selective angiography of the right internal iliac artery showed no contralateral source of haemorrhage. Twenty-four hours following embolisation, the patient underwent a contrast CT scan of the abdomen and pelvis. This demonstrated the embolisation coils in the left uterine artery remnant, no intra- or extra-peritoneal haemorrhage or collection and no abnormal contrast collection in the upper vagina.

She was discharged home 2 days post-embolisation on oral antibiotics and iron with haemoglobin of 8.8 g/dl. Four weeks post-discharge, she had a normal pelvic ultrasound scan; she was no longer anaemic; and on the 8 week followup, she remained asymptomatic and she was discharged from our care.

Discussion

In contrast to true aneurysms, pseudoaneurysms are not surrounded by the normal three vascular wall layers of tunica intima, media, and adventitia. They are bordered by adjacent perivascular tissue and thrombus, creating a pseudocapsule. They can undergo spontaneous thrombosis and resolution, or become infected and develop into a septic pelvic collection. Alternatively, they can cause pain and pressure symptoms to nearby organs. Occasionally, they can be asymptomatic and present as an incidental finding on routine post-operative imaging [2]. Spontaneous rupture is the most serious complication with high morbidity and mortality. Depending on the cause of the pseudoaneurysm and the type of the antecedent operation, rupture presents with intra- or extra-peritoneal bleeding, uncontrolled metrorrhagia or vaginal vault bleeding.

Pelvic surgery is the most common cause of uterine artery pseudoaneurysms. In obstetrics, they can present with secondary postpartum haemorrhage after caesarean section [3, 4]; in gynaecology, they can develop post-hysterectomy, myomectomy or dilatation and curettage [1, 5–9].

The natural history involves injury to a vessel during dissection or suturing, incomplete vascular sealing or extensive desiccation which results in the development of haematoma. Central liquefaction of the haematoma leaves a cavity which communicates with the parent vessel [2]. Rupture is unpredictable and characteristically delayed, as time is needed for this process to take place and the pseudoaneurysm to form. In our case, although there were no intra-operative difficulties with haemostasis of the uterine vessels, extensive desiccation of the uterine pedicles and the surrounding connective tissue might have resulted in perivascular inflammation therefore contributing to the development of the pseudoaneurysm.

Diagnosis is suspected from the clinical history and confirmed with imaging investigations. Ultrasound scan is usually the first line investigation and shows a characteristic well-defined hypoechoic structure with central turbulent blood flow at Doppler analysis. Blood enters the pseudoaneurysm during systole, and then during diastole, the drop of the pressure within the neck of the pseudoaneurysm results in reversal of flow [2, 6]. Arteriography is the gold standard diagnostic investigation [10]. In our case, on initial presentation, ultrasound scan was not performed as there was heavy bleeding and it was thought that imaging would delay treatment. In our case, if an ultrasound scan had been performed, the diagnosis of a pseudoaneurysm might have been considered and uterine artery embolisation performed earlier, thus avoiding surgery.

It can be argued that re-suturing of the vault could have been responsible for the formation of the pseudoaneurysm. However in our case, the clinical presentation was delayed torrential vaginal bleeding which is characteristic for a pseudoaneurysm of the uterine artery. Therefore, we believe that the pseudoaneurysm was the cause of bleeding, and suturing and vaginal packing only temporarily arrested it.

Uterine artery embolisation is the treatment of choice with very good success rate in occluding the affected vessel. Surgery should be avoided as it can delay definite treatment and result in further bleeding and inadvertent ureteral and/or bladder injury caused by the application of multiple stitches in a recently operated pelvis.

Until now, few cases of uterine artery pseudoaneurysm post-gynaecological surgery have been reported in the literature. To our knowledge, this is the first time this is reported after laparoscopic hysterectomy. However, we believe that this complication is not related specifically to the laparoscopic approach as similar cases have been reported after open or vaginal hysterectomy [6, 8].

It is unclear to what extent this complication was related to the operative technique and whether it could have been prevented. Extensive electrosurgery of the vascular uterine pedicles and post-coagulation inflammation could have contributed to the pseudoaneurysm formation. We do not believe that the use of conventional bipolar energy instead of advanced bipolar sealing device would have made any difference in preventing this complication. What is more important than the actual device used is to carefully skeletalise the uterine vessels before coagulation. This might reduce the risk of a pseudoaneurysm formation by reducing the incorporation of extra connective tissue in the vascular bundle that is coagulated. This could in turn, minimise tissue necrosis and perivascular inflammation and potentially prevent a pseudoaneurysm.

Conclusion

Uterine artery pseudoaneurysm should be considered whenever delayed heavy vaginal bleeding occur postlaparoscopic hysterectomy. Although the clinical presentation resembles that of a vaginal vault haematoma, it is the severity of bleeding that should raise the suspicion of a pseudoaneurysm. These patients are usually in haemorrhagic shock and the correct diagnosis should be prompt. This is essential as embolisation is the treatment of choice rather than surgery. Surgery can result in delaying definite treatment of this life-threatening condition.

Conflict of interest The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

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