YOUR GUESS IS AS GOOD AS MINE; DEALING WITH RECURRENT SECONDARY POST-PARTUM HAEMORRHAGE CAUSED BY UTERINE ARTERY PSEUDOANEURYSM

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ABSTRACT Uterine artery pseudoaneurysm (UAP) is a rare cause of secondary postpartum haemorrhage (PPH). Ultrasound may suggest this condition, and pelvic arteriography is done to confirm the diagnosis and to institute treatment via uterine artery embolization. Herein, we describe a case who experienced recurrent episodes of secondary PPH with inconclusive findings on ultrasound and pelvic arteriography. Foley catheter balloon was used several times to arrest acute bleeding episodes. Exploratory laparotomy revealed pseudoaneurysm of the branch of the right uterine artery. Transection and ligation of the involved artery followed by ligation of bilateral internal iliac arteries were performed. Post-operation, the patient made a full recovery. This case highlights uterine artery pseudoaneurysm as a rare but important cause for secondary PPH. We recommend a simple measure to provide intrauterine tamponade by using a Foley catheter balloon. Diagnosis of UAP may not be straightforward, and this condition may only be discovered during surgery.

KEYWORDS secondary PPH, pseudoaneurysm, pelvic angiography, intrauterine tamponade, Foley catheter balloon

Case report

A 26-year-old, para 2, was admitted with massive per-vaginal bleeding 28 days after caesarean delivery for foetal distress. The caesarean section was uncomplicated with a total blood loss of 800mls. The immediate and early post-partum period were uneventful. Clinically she was pale, with BP of 89/50 mmHg and pulse rate of 120 beats-per-minute. The uterus was not palpable, the vaginal examination was unremarkable, and the active bleeding had stopped spontaneously. On-site haemoglobin level was 7.5gm/L. The transabdominal scan shows normal size uterus with a thin endometrium. She was diagnosed as secondary PPH with possible endometritis in hypovolaemic shock. Resuscitation with intravenous fluids and blood and treatment with antifibrinolytics and antibiotics were instituted. She was discharged home after three days free of bleeding.

However, this was just the beginning of everyone's nightmare. She subsequently experienced five more episodes of significant uterine bleeding over the next 50 days, as illustrated in table 1. Each episode of bleeding occurred abruptly without any apparent precipitating cause. The bleeding stopped spontaneously in the first two admissions and subsequently in response to intrauterine balloon tamponade.

Suction and curettage were eventful as massive acute bleeding occurred with 1.8litres of blood loss. Intrauterine tamponade provided by Foley catheter balloon managed to stop the acute bleeding instantaneously. Retained gestational products, molar pregnancy and coagulopathies were ruled out by negative histopathology, normal serum beta HCG and normal blood works. Possibility of rare causes of secondary PPH, including

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Table 1 Summary of multiple ward admissions for recurrent secondary PPH.

Admission, Post-partum Day	Clinical presentation	Haemoglobin (Hb)	Management
1^{st}, Day 28	Massive vaginal bleeding, hypovolaemic shock	Hb: 7.5g/L	-Resuscitation -Antibiotics, antifibrinolytics
2^{nd}, Day 38	Heavy vaginal bleeding.	Hb: 8.5g/L	-Another course of antibiotics in view of doubtful compliance to previous treatment
3^{rd}, Day 45	Heavy vaginal bleeding.	Hb: 7g/L	-Suction and curettage complicated with bleeding: 1.8 litres. -Bleeding stopped after intrauterine tamponade -Beta hCG: <0.1mIU/L -HPE: necrotic tissue, no evidence of trophoblast.
4^{th}, Day 65	Massive vaginal bleeding, hypovolaemic shock	Hb: 5g/L	-Transfusion: 4 pints blood and blood products. -Bleeding stopped after intrauterine balloon tamponade. -Pelvic angiogram: No vascular abnormality. Uterine arteries embolized with Gelfoam
5^{th}, Day 80	Increased vaginal bleeding	Hb: 10 g/dL	-Bleeding stopped after intrauterine balloon tamponade.
6^{th}, Day 87	Increased vaginal bleeding	Hb: 10 g/dL	- Exploratory laparotomy: day 90 post- delivery after massive 1.5 litres bleeding while in ward

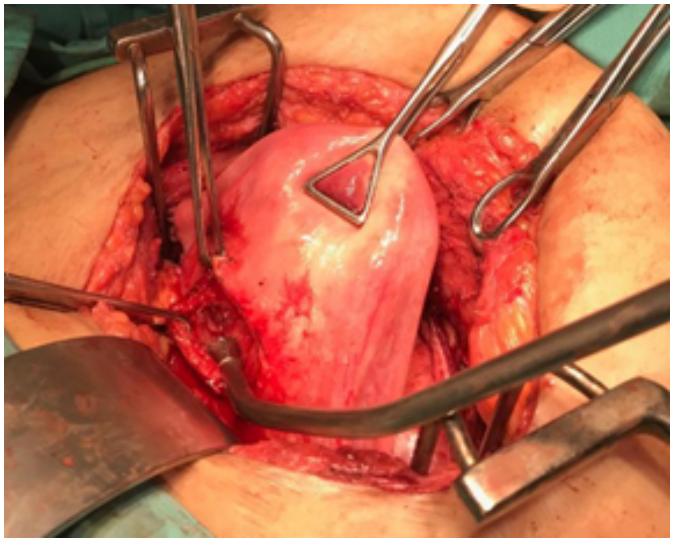

Figure 1: Demonstrating right uterine artery pseudoaneurysm.

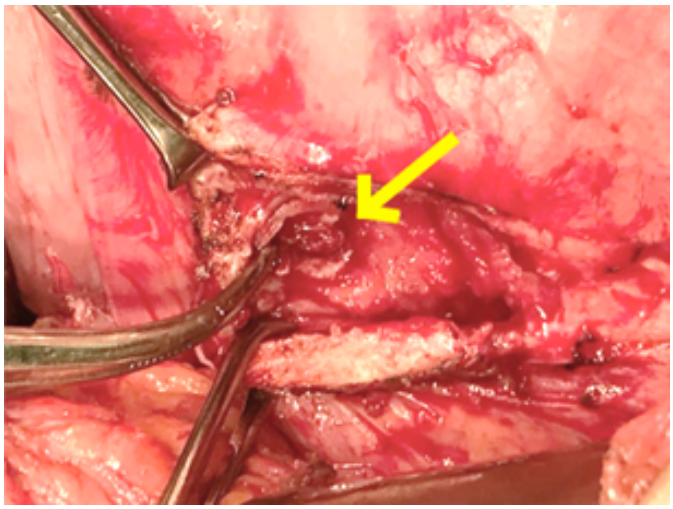

Figure 2: Close up view of right uterine artery pseudoaneurysm showing a 'pocketlike' cavity (arrow) in the right posterior endometrium, prominent branch of right uterine artery was 'inside' the cavity (artery forceps).

uterine vascular anomalies, was entertained. Both B-mode and Doppler ultrasound, however, were not suggestive of this diagnosis. The patient had refused surgery because of possible hysterectomy. She was then referred to Hospital Kuala Lumpur which is located 450km away for pelvic arteriography. A pelvic angiogram revealed no evidence of any vascular abnormalities. Despite this, bilateral uterine artery embolization was carried out in view of her significant clinical presentations. Unfortunately, she experienced another two more episodes of bleeding which were controlled by intrauterine tamponade and on day 90 post-partum, she finally agreed for surgical intervention.

Intraoperatively, a normal involuted uterus was seen without evidence of scar dehiscence. The previous uterine incision scar was opened until the endometrial cavity. There was a 1.5cm x 1cm, a small pocket-like cavity at the right posterior endometrium which contained a prominent branch of right uterine artery and blood suggestive of pseudoaneurysm (figure 1 and figure 2). The rest of endometrial cavity was explored and was found to be normal. The affected branch of the right uterine artery transected and ligated followed by ligation of bilateral internal iliac arteries. She made an excellent full recovery post-operation.

Discussion

Post-partum haemorrhage (PPH) remains one of the leading causes of maternal mortality worldwide causing 27.1% of the direct maternal death [1]. A large majority of PPH cases are categorized as primary PPH. In contrast, secondary PPH, which is less common than primary PPH, is defined as any major bleeding occurring 24 hours to 6-12 weeks after delivery [2]. The average incidence of secondary PPH in developed countries is around 1% while the incidence of severe secondary PPH after caesarean section is much lower, approximately about 0.1% [3]. Despite the low incidence, secondary PPH is not only a significant cause of maternal morbidity but also a potential cause of mortality following childbirth.

Common causes of secondary PPH include retained gestational products, subinvolution of the placental bed, and endometritis. Rare causes include arteriovenous malformations, pseudoaneurysm of uterine artery, scar dehiscence and choriocarcinoma [4]. We considered all these possibilities in the management of this patient. The final diagnosis in our case was uterine artery pseudoaneurysm (UAP) based on the operative

findings. A pseudoaneurysm is a blood-filled cavity, with turbulent flow, that communicates with the arterial lumen. It results from an inadequate sealing of a lacerated artery as a consequence of surgery, penetrating trauma, neoplasm or infection [5] and in our case, most probably related to the previous caesarean section. The pseudoaneurysm margins are formed by the thrombus originating from the lacerated artery and are not surrounded by three arterial layers as in a case of the true aneurysm. The incidence of UAP in child-bearing females is estimated at 0.2 %–0.3% [6].

The nature of the patient's bleeding was abrupt, short-lived and responded well to intrauterine tamponade; these features were all suggestive of vascular anomalies of the uterus. Rupture of a pseudoaneurysm can cause severe haemorrhage. In some cases, the rupture is limited by the surrounding tissues, causing intermittent bleeding [7], which was the prominent nature of our patient's presentation. Diagnosis of pseudoaneurysm could be suspected initially based on ultrasound findings. B-mode ultrasound may show pulsatile anechoic or hypoechoic lesion within the myometrium. With colour doppler ultrasound, the lesion gives colour signals with aliasing at its neck due to turbulent blood flow [8]. The characteristic to-and-fro pattern on spectral doppler has been reported to have 95% of diagnostic sensitivity in cases of UAP [9,10]. In cases where colour Doppler and grey-scale ultrasound findings are equivocal, pelvic angiography is used to confirm this diagnosis [8]. In our case, however, all these diagnostic findings were not evident. Diagnosis of UAP may be hindered by few major pitfalls, i.e. spontaneous hemostasis with delayed recurrence of bleeding and absence of typical ultrasound features at admission due to hemorrhagic shock and the small diameter of the lesion [11]. Cessation of bleeding with stable blood clots forming at the site of pseudoaneurysm and small UAP size could be the reasons for inconclusive findings during diagnostic tests in our case.

The role for pelvic angiography, however, is not limited to diagnostic purpose only but also for treatment purpose. In our case, despite negative findings at angiography, uterine artery embolization (UAE) was carried out, given her significant clinical history. UAE however, is generally less efficacious in secondary PPH as compared to primary PPH (77% versus 93%) [12]. On the other hand, in one case series, arterial embolization allowed controlling the bleeding in 94% of patients after one embolization session [13]. Inadequate embolization due to extrauterine feeding arteries [4] and the inability to perform super-selective embolization in apparently 'normal angiography', are possible management obstacles in our case. The patient was embolized with Gelfoam, an agent which is highly hemostatic with low ischaemic complications but provides only temporary occlusion. Gelfoam, however, has been shown to be an effective treatment for ruptured pseudoaneurysm in other reports [13,14]. All the above factors could have caused embolization less effective in the management of our case.

The choice of treatment may also depend on the specific resources available in each institution. Although angiographic embolization is a safe and effective procedure for treating postpartum haemorrhage resulting from UAP in hemodynamically stable patients, uterine artery ligation may be the surgical procedure of choice for hemodynamically unstable patients when fertility preservation is desired [7]. Hysterectomy is one of the surgical options when the preservation of fertility is not important. Direct thrombin injection under ultrasound guidance has also been described as a treatment for UAP [15]. This is an attractive option because it does not require surgery or sophisticated radiological equipment; however, its indications and effectiveness have not yet been determined [15]. In the beginning, our patient had refused surgery due to hysterectomy risk since she wants to preserve her fertility. Consent was only given after we have exhausted our treatment options.

Secondary PPH is rarely managed by intrauterine tamponade because most cases are non-vascular causes, e.g. retained tissues or endometritis, which are easily treated with uterine curettage or antibiotics. Bakri balloon has been reported to successfully manage a case of secondary PPH in a sub-involuted uterus. However, the author cautioned against using this technique, especially after caesarean section and where there is severe sepsis with organ failure [16]. Our patient presented with massive uterine bleeding almost a month from the delivery date when the uterus has involuted to its normal size, and Bakri balloon is not suitable in this case because of its size. We used Foley's balloon catheter several times, in this case, to halt the acute uterine bleeding successfully. This strategy managed to buy us time to resuscitate adequately and even to stabilize the patient to be transported away to another centre. We recommend this strategy to be used in secondary PPH cases suspected to be caused by uterine vascular abnormalities as a temporary measure during the acute bleeding episode.

Conclusion

Uterine artery pseudoaneurysm is rare but, should be listed as a possible cause of postpartum haemorrhage, especially after caesarean section. Intrauterine tamponade with a Foley catheter balloon is a simple measure that can be used to control acute bleeding episode in such a case. Inconclusive diagnostic tests are uncommon, and a high degree of suspicion is needed to diagnose this condition, particularly when one is contemplating fertility-sparing surgery.

Conflict of interest

There are no conflicts of interest to declare by any of the authors of this study.

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