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Failure of Coil Embolization in Secondary Postpartum Hemorrhage: A Case Report

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Abstract :- Postpartum haemorrhage (PPH) is a life-threatening condition and remains a leading cause of maternal mortality. Embolization is an effective therapeutic strategy for PPH with the advantages of being fast, repeatable, and the possibility of fertility preservation. A young 30 year old female P2L2 presented to our department with secondary postpartum haemorrhage initially managed conservatively followed by coil embolization and finally hysterectomy after the failure of conservative and embolization procedure.

Keywords:- *Postpartum haemorrhage, Embolization*

I. INTRODUCTION

The definition of postpartum haemorrhage is blood loss of >500 mL after normal vaginal delivery and >1000 mL after cesarean section.[1]This was redefined in 2017 by the American College of Obstetrics and Gynaecology as a cumulative blood loss greater than 1000 mL with signs and symptoms of hypovolemia within 24 hours of the birth process, regardless of the route of delivery. While this change was made with the knowledge that blood loss at the time of delivery is routinely underestimated, blood loss at the time of vaginal delivery greater than 500 mL should be considered abnormal with the potential need for intervention.[2 3 4]Postpartum haemorrhage can be primary that occurs with first 24 hours of delivery or secondary that occurs after 24 hours and up to 12 weeks of delivery.[5]

Conservative management of PPH includes resuscitation, blood transfusion, and administration of uterotonic drugs such as oxytocin and prostaglandin, uterine compression and intrauterine balloon tamponade. When conservative management fails, TAE or surgical management is implemented without any delay. Surgical management includes vessel ligation [bilateral ligation of uterine or internal iliac arteries], uterine compression suture and hysterectomy[6].

It is widely accepted that TAE is an effective therapeutic strategy for PPH of various causes[7]. TAE has the advantage of being a fast, repeatable procedure and can be performed without general anaesthesia. Moreover, TAE preserves the uterus and makes future fertility possible.1

The incidence of secondary PPH has been reported to be 0.2 to 0.8% and it is one the most common indications for readmission after delivery.[8-11] Although the number of women affected by secondary PPH is relatively small compared to number of women affected by primary PPH, it can cause serious complications if the diagnosis and appropriate treatments are delayed. Common causes of secondary PPH include retention of placenta, endometritis and delayed placental bed involution[10, 11]. Other less common etiologies are congenitalcoagulopathies, placenta adherens, caesarean scar dehiscence, uterine pseudoaneurysms, and uterine rupture.[11 12 13 14]

Initial management is aimed at achieving hemodynamic stabilisation. Subsequently, specific managements depend on the cause of bleeding. In most cases, infection is treated with uterine evacuation if retained placental tissue is suspected. In certain circumstances such as continuous bleeding, uterine perforation and uterine pseudoaneurysms, hysterectomy or arterial embolization may be indicated.[8-10, 15]

II. CASE PRESENTATION

A thirty, year old female, P_2 L_2, first delivery was normal vaginal delivery and second was done by LSCS on 26th march 2022 at private hospital, presented with complaint of secondary postpartum haemorrhage on 6th may 2022 at our hospital. On admission, vitals of patient were stable with blood pressure of 110/70, pulse rate of 84 b/m and spO_2 of 95% on room air. Baseline investigations were done, haemoglobin was 5.6gm/dL, WBC - 8.7k and platelet count was 196k. Beta hcG was negative. First radiological investigation done was USG abdomen and pelvis [done on 6th may] with findings as:

- Hepato-biliary system – normal
- Post-delivery normal uterus with endometrial thickness as 5mm
- No significant post-surgical collection seen on USG.

Management of patient was started with antibiotics, oxytocin drip, misoprostol 800 mcg in posterior fornix and later first blood transfusion on 7th may. Simultaneously CECT abdomen and pelvis was planned and was done at next at SMHS hospital Srinagar. CECT findings were as:

Heterogenously enhancing material noted in intrauterine cavity with hyper-vascular content in left cornu measuring 21x22mm. Endo-cervical canal is dilated.

Impression on CECT was given as retained products of placenta. Then check curette was done on 12th may. Significant products were removed, complete haemostasis was achieved at the procedure. Products which were removed were sent for histo-pathological examination

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[second blood transfusion was given during check curette]. Postoperative period of patient was uneventful, vitals were stable, haemoglobin was 8gm/dL, WBC – 5.9k and platelet count was 360k.

She was advised to come for follow up after 2 weeks with histo-pathological report; however, she reported to hospital on emergency basis 12 days later with complaint of bleeding per vagina again.

On admission, patient was concious and oriented but was pale, pulse rate was 120 b/m, bp 140/94 and spO_2 of 97% on room air. Per abdomen examination of patient showed non tender and non-distended abdomen, on per vaginal examination around 20 to 30 cc clots were removed with no active bleeding. Haemoglobin was 7.3gm/gL, WBC 10.5k, platelet count was 280k, INR 1.07, PT-12.9, aPTT 18.9 second.

USG abdomen and pelvis was done [24th may] with findings as:

- Liver, gallbladder, pancreas, bilateral kidneys were normal.
- Uterus was ante-verted with normal myometrium except for few clots at suture line, thin rim of fluid in endometrial cavity, bilateral adnexa normal, No abdominal or pelvic collection seen at time of scan.

This time patient was managed conservatively by giving iv antibiotics [piperacillin and tazobactam] and 1gm of ferric carboxymaltose was given on 26th may 22. She was started on norethisterone 10mg daily and was discharged on same on 30th may 22. On her routine follow up, she presented with histo-pathological report of curettage procedure done in previous admission with findings as:

Sections examined two fragments, one fragment composed of endometrium in secretory phase and other fragment comprises of endocervical tissue with chronic inflammation. No specific pathology seen in sections examined.

3 weeks later she presented again with complaint of bleeding per vagina, on general physical examination of patient, she was concious and oriented, pulse rate was 140 b/m, spO2 96% on room air, BP was 110/76, abdomen was soft and non-tender and non-distended on examination. On per vaginal examination 1 litre of clots were removed from vagina but there was not any active bleeding. Haemoglobin was 5.7 gm/dL [significant fall], WBC 3.3k and platelet count was 247k. LFT and KFT was within normal limits, coagulogram was normal, stool for occult blood was negative, urine for cast`s negative, serum ferritin was 269.42. TVS findings were as below:

- Endometrial canal clear, myometrium echotexture normal, uterus having normal vascularity, no arterio-venous malformation was seen and endometrial thickness was 2mm. Pelvic CT angiography was done with findings as below:
- Left uterine artery shows evidence of 8.5x7x9mm pseudo-aneurysm formation in the region of lower uterine segment on left side. Right uterine artery is unremarkable for any pseudo-aneurysm. Uterus is mildly bulky with evidence of non-enhancing contents within the cavity likely minimal RPOCs.

Interventional radiology department of SMHS was contacted and they advised embolization of bleeding vessel as the favourable option for the patient keeping in view her young age, parity only two and considering the less invasive nature of the procedure with good efficacy. Meanwhile patient received three blood transfusions on haemoglobin of 5.7gm/dL while she was being prepared for embolization. She was sent for uterine artery embolization on 2nd July 22.

Procedure: under all aseptic precautions, left common femoral artery punctured with 18 G puncture needle and 6 F secured in place. Using 5F Picards catheter left common iliac artery catheterised, left internal iliac artery run taken which showed pseudo-aneurysm from left uterine artery. Selective catheterisation of left uterine artery done using progreat micro catheter. Catheter advanced into tortuous uterine artery up to the level of pseudo-aneurysm. Four 18-2-2 coils deployed in left uterine artery.

Repeat left uterine artery angiogram reveals complete occlusion of artery and no filling of aneurysm. Catheter removed and haemostasis achieved. Patient was advised limb immobilisation for 8 hours. Post-embolization period of patient remained satisfactory, remained stable haemodynamically, afebrile with no complaint of bleeding per vagina or bleeding/hematoma formation at puncture site. Patient was then discharged from hospital on 7th July.

4th time on 31st July, she presented with complaint of heavy bleeding per vagina. On her per vaginal examination, vagina was full of clots, bleeding per vagina was significant. Haemoglobin was 6.4gm/dL, WBC 12k, platelet count was 344k. LFT KFT normal, blood sugar random 90mg/dL, beta hcg was negative, coagulogram normal and TSH was 1.2.

USG pelvis (TVS) [on first august] with findings as:

- Uterus shows small anechoic area within myometrium measuring 23x16mm showing minimal vascularity on color Doppler. Echogenic area measuring 13x12mm seen within endometrial cavity with feeding vessel sign suggestive of endometrial polyp.
- Repeat CT pelvic angiography was done on 2nd august with findings as below:
- Evidence of focal contrast out-pouching measuring 41x28x36mm is seem along the course of left uterine artery along left lateral wall of uterus suggestive of pseudo-aneurysm. Few metallic density areas are seen along left uterine artery suggestive of surgical clips.

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Fig. 1: Axial CT angiography showing well defined contrast enhancing pseudo-aneurysm arising from left uterine artery. Previous aneurysmal clip seen in proximal left uterine artery

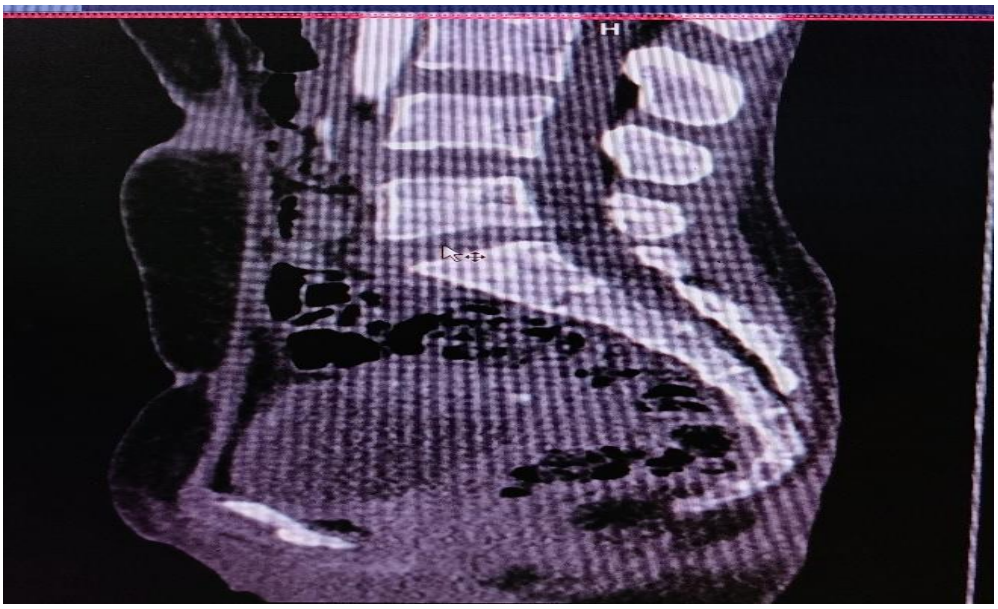

Fig. 2: Sagittal CT angiography image of the same patient showing pseudo-aneurysm

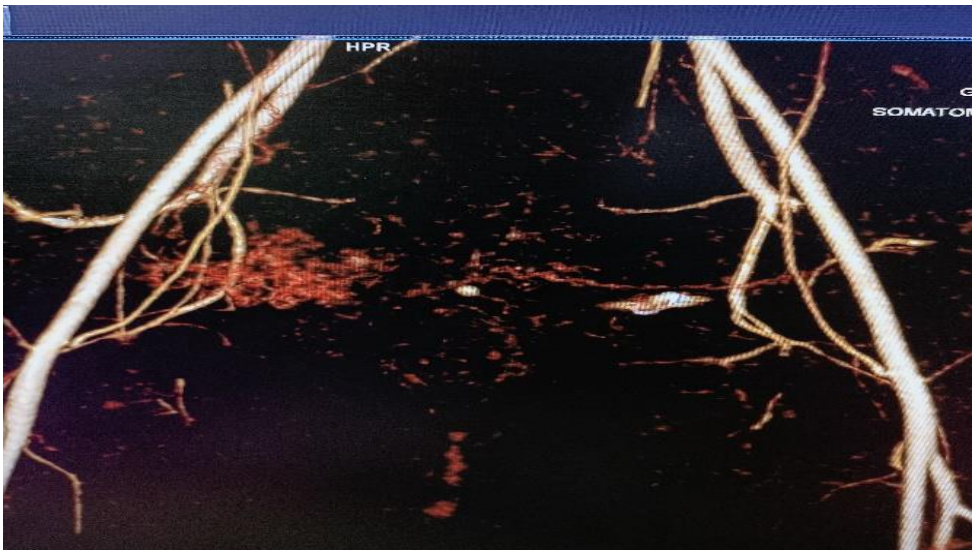

Fig. 3: VRT image showing left uterine artery pseudo-aneurysm. Previous aneurysmal clip also seen in left uterine artery

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Patient was planned for hysteroscopy and polypectomy but she did not give consent for the same, she instead insisted for hysterectomy despite negative counselling. In the ward while being built up, patient started bleeding profusely and almost went into shock and she was taken for emergency laparotomy after taking the proper informed consent. Operation performed was total abdominal hysterectomy and bilateral salpingectomy with preservation of bilateral ovaries.

Intraop findings were:

- Tortuous vessel felt on left side below the lower uterine segment.
- Uterine surface apparently normal.
- Bilateral tubes and ovaries normal.
- POD normal apparently normal.
- Bladder firmly adherent to lower uterine segment

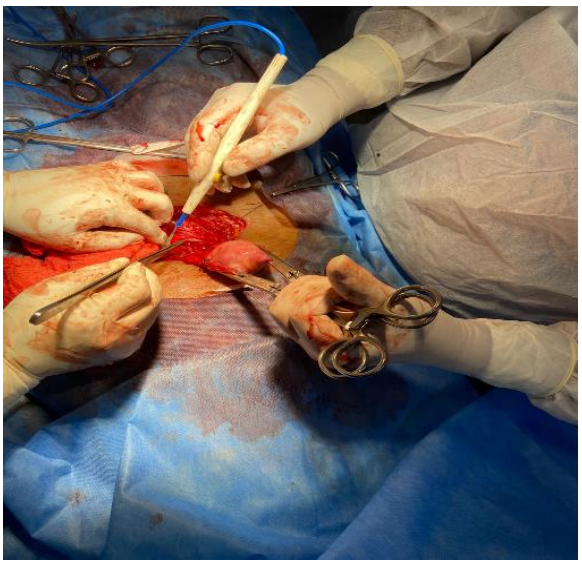

Fig. 4: Image showing the dissection being done at the level of cervico-uterine junction to preserve cervix

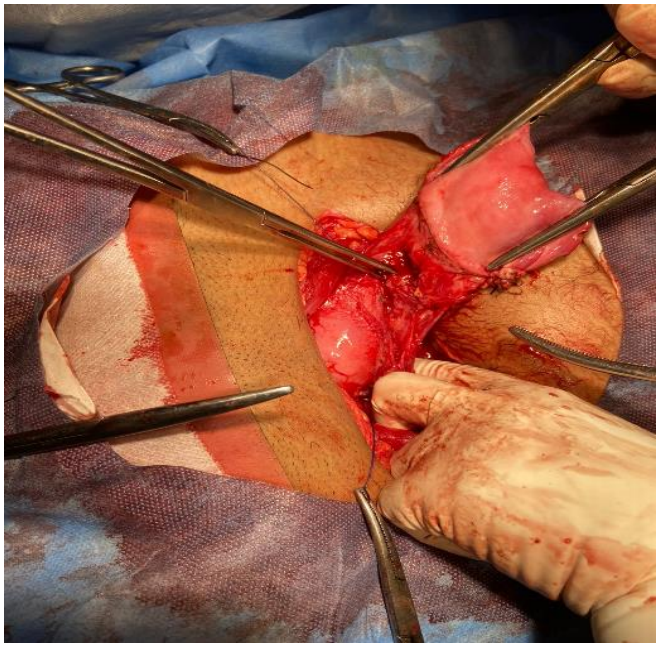

Fig. 5: Image showing the small tortuous bleeders at the cervico-uterine level being ligated

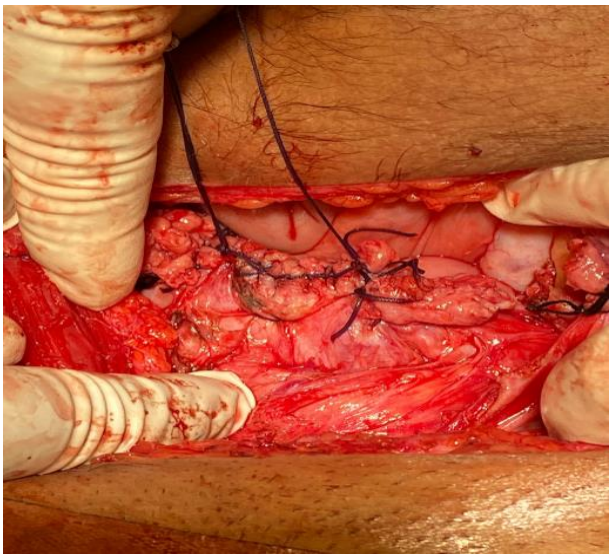

Fig. 6: Image showing the vaginal cuff closure

Attempts were made to preserve the cervix but was not possible because of tortuous vasculature which was noted on the left side of uterus in lower segment, on cutting open the uterus, cavity was normal and except for tortuous vessel on left side utero-cervical junction, no other abnormality was found. Two blood transfusions were given intra-op followed by one more blood transfusion and 1.5gm ferric carboxymaltose in post-operative period. Finally, the patient was discharged on 29th of august with vitals stable and haemoglobin of 9gm/dL.

III. DISCUSSION

An arterial pseudo-aneurysm is a false aneurysm caused by trauma to arterial wall. Compared to true arterial aneurysm, pseudo-aneurysm do not contain all three layers of arterial wall [adventitia, media and intima]. They are blood filled cavities that vary in size and communicate with the lumen of their parent artery. When the arterial wall is injured, contact will be maintained with the parent vessel and extravasated blood will dissect through the arterial tissues. For this reason, obstetricians and gynaecologists will often identify pseudo-aneurysms after instrumentation of uterus during dilation and curettage, myomectomy, operative assisted deliveries or caesarean sections. In the literature, it is estimated that post caesarean section accounts for 47.4% of all cases of uterine artery pseudo-aneurysm, making caesarean delivery most common cause of uterine artery pseudo-aneurysm[16]. As postoperative and post-delivery imaging is not routinely performed, it is difficult to estimate how many cases of uterine artery pseudo-aneurysms without vaginal bleeding go unnoticed.

The diagnostic imaging modality of choice is transvaginal ultrasonography with colour doppler. On ultrasound doppler, a “to and fro” pattern may be visualised within a hypoechoic mass in the neck of pseudo-aneurysm. The arterial blood will flow into aneurysm cavity during systole and then reverse into the parent artery during diastole due to pressure gradient. Other imaging modalities that can be diagnostic are MRI and CT with contrast.

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There are various treatment approaches to the management of uterine artery aneurysm, the patient's preferences for future fertility should guide care in the shared decision-making process. For patients who are haemodynamically stable and desire fertility sparing therapy, angiography is the gold standard as it is both diagnostic and therapeutic. Interventional radiological uterine artery embolization is a highly effective technique for controlling acute and chronic uterine bleeding. Benefits of this procedure include low complication rates, avoidance of surgical risks and fertility preservation. Haemodynamically stable patients who desire fertility sparing therapies should be offered interventional radiological uterine artery embolization prior to invasive surgical management in non-malignant pelvic bleeding. Glue, polyvinyl alcohol particles, gelfoam and micro-coils are among the particles that can be used in interventional radiological embolization procedures with the most of these materials used being polyvinyl alcohol particles.

For patients who need emergent treatment, as in those who are haemodynamically instable, hysterectomy, removal of pseudo-aneurysm, uterine vessel coagulation or ligation are viable options but may affect or eliminate patient ability to conceive.

Precise knowledge of normal and varying anatomy of female genital tract is essential for accurate interpretation of angiographic images and safe performance of PPH embolization. The internal iliac artery divides into two divisions. The anterior division of internal iliac artery is subject to numerous variations and gives rise to umblical, superior vesical, obturator, inferior vesical, vaginal, middle rectal, internal pudendal, inferior gluteal and uterine arteries. The posterior division of internal iliac artery gives to iliolumbar, lateral sacral and superior gluteal arteries.

The uterine artery shows characteristic U-shaped course and consists of descending or parietal segment, transverse or arch segment and ascending or marginal segment which runs along the uterine side and has numerous intramural branches [arcuate arteries]. The cervico-vaginal branch arises from transverse segment and supplies blood to cervix and vagina[17.] Ovarian and round ligament arteries often participate in a complex anastomotic network that provides blood supply to uterus[18]. The ovarian arteries arise from abdominal aorta below the renal artery, and round ligament artery arises from inferior epigastric artery[18]. Collaterals to uterus from ovarian artery, inferior mesenteric artery, round ligament artery, and internal pudendal artery are well known and can be a bleeding source in PPH[17 19].

The general rule regarding the embolization of bleeding arteries is:

- Embolization of bilateral uterine arteries regardless of the presence of active bleeding foci.
- Embolization of anterior division of bilateral internal iliac arteries in case the uterine artery could not be easily accessed, e.g. in patients with hysterectomy or hemodynamic instability or when bilateral uterine artery embolization fails to stop bleeding.
- Embolization of other potential bleeders based on additional angiography or aortography if bilateral uterine and internal iliac artery embolization fails. The anastomotic arteries especially the ovarian artery and round ligament have an important role in re-bleeding.

Possible anastomotic arteries causing PPH other than uterine artery:

- Ovarian artery
- Round ligament artery of uterus
- Obturator artery
- External pudendal artery from femoral artery
- Deep circumflex iliac artery
- Superior rectal artery from inferior mesenteric artery
- Middle rectal artery
- Median sacral artery
- Iliolumbar artery

The re-bleeding rate after TAE has been reported to range from 5.2% to 13.5% despite its technical success[22 23 24]. The main reason for re-bleeding after TAE is recanalization of embolized arteries followed by opening of collateral communications[17 19]. In addition to confirmation of uterine artery recanalization, search for presence of spontaneous anastomosis, such as round ligament artery, ovarian artery, middle rectal artery or inferior mesenteric artery is necessary on the angiography in search of re-bleeding.[20]

Repeated TAE is safe and effective for managing re-bleeding after a single session of TAE.[20 22]

The choice of embolic materials depends on the combination of angiographic findings, achievable catheter position and the operator preference.

Gelatin sponge particles are the most commonly used primary embolic material for PPH embolization. Gelatin sponge particles are mixed with diluted contrast medium to produce a slurry, which is subsequently injected into arteries until stasis or occlusion of blood flow is evident during angiography[25]. They enable temporary occlusion for 3 to 6 weeks and recanalization of target arteries which is advantageous for future fertility[7]. However, TAE with gelatin sponges as the sole embolic material in patients with coagulopathy has been reported to limit the effectiveness in achieving the successful haemostasis[26]. In addition, in patients with active bleeding, such as patients with pseudo-aneurysm, use of gelatin sponge may be inadequate for effective treatment[27 28]. The use of N-butyl cyanoacrylate [NBCA] should be considered in patients with:

- Active bleeding that is contrast extravasation of pseudo-aneurysm.
- Haemodynamically unstable conditions.
- Failed embolization with gelatin sponge particles [usually at the repeated session of embolization].

The mechanism of NBCA embolization involves rapid polymerisation by making contact with blood and complete blocking of arterial injury or filling the pseudo-aneurysm and thus providing effective embolization regardless of coagulopathy. Therefore, emerging TAE using NBCA can

be a beneficial first choice treatment for PPH with DIC, extravasation or pseudo-aneurysm.[27 29]

The use of polyvinyl alcohol particles is not recommended as they may cause uterine necrosis.[30]

Metallic coils can be used to occlude ruptured pseudo-aneurysms as the sole or as adjunctive embolic material.[27]

IV. CONCLUSION

Postpartum haemorrhage be it primary or secondary can be life threatening, although primary is more associated with urgency and maternal mortality. However, patients with secondary postpartum haemorrhage are haemodynamically stable and they usually require less number of blood transfusions. Although pseudo-aneurysm of the uterine artery is known to be a rare cause of delayed postpartum haemorrhage, it is highly associated with secondary PPH in many patients. Trans-arterial embolization should be considered after conservative management fails in postpartum haemorrhage due to its unique characteristics of being fast, highly efficacious, wide indication, minimal invasiveness, and uterine preservation.

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