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A rare cause of secondary postpartum haemorrhage

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Abstract
Secondary postpartum haemorrhage (PPH) after caesarean section due to uterine artery pseudoaneurysm is an unusual complication. We report a case of 26 years-old female (P2L2) who had lower segment caesarean section (LSCS) was presented after 36th day with excessive bleeding per vagina for 1 day. She was diagnosed to have pseudoaneurysm by CT angiography. Selective transcatheter arterial embolization of pseudoaneurysm was planned and digital substraction angiography was done. Left iliac arteriogram revealed a pseudoaneurysm from the terminal part of the left uterine artery. Post-operative period was uneventful and the bleeding stopped. In conclusion, pseudoaneurysm of uterine artery should be considered as a differential diagnosis, when the patient presents with secondary PPH after caesarean section. Embolisation is the treatment of choice and its prevents a hysterectomy.

Keywords: Pseudoaneurysm, angiography, embolization, uterine artery, hysterectomy

Introduction
Postpartum haemorrhage (PPH) is a complication of delivery and the most common cause of maternal death, accounting for about 35% of all maternal deaths worldwide. PPH is commonly defined as any blood loss>500 ml following vaginal delivery and >1000 ml after cesarean section. Definitions vary, however, and diagnosis of PPH is subjective and often based on inaccurate estimates of blood loss [1]. PPH is often classified as primary, occurring within 24 hours of birth and is more common form of PPH [2] or secondary defined as bleeding in excess of normal lochia after 24 hours and 6-12 weeks postpartum [3]. The secondary PPH is rare and after caesarean section occurs in about 1: 365 cases [4]. Overall incidence of secondary PPH in developed world varies from 0.47% to 1.44% [5].
The rare causes of secondary PPH are uterine artery pseudoaneurysm (UAP), AV malformations, injured vessel and choriocarcinoma. Pseudoaneurysm (PA) of the uterine artery is an extraluminal collection of blood with turbulent flow communicating with the lumen of the parent vessel through a defect caused by trauma, and when connected with the uterine cavity, it causes recurrent hemorrhage [6]. Proper treatment requires an accurate diagnosis, which is generally based on the results of color Doppler sonography and confirmed by performing uterine artery angiography. Uterine artery embolization should be considered as the treatment of choice for haemodynamically stable patients [7]. Herein we report a case of secondary post-partum hemorrhage in a 26 years old P2L2 woman 36th days after cesarean section due to pseudoaneurysm of uterine artery which was managed successfully with uterine artery embolisation.

Case Report
A 26 years-old female (P2L2) who had lower segment caesarean section (LSCS) was presented after 36th day with excessive bleeding per vagina for 1 day. On general examination, she was moderately built and having pallor+++, afebrile, pulse rate 110/min, BP 80/50mmHg, per abdominally healthy suprapubic scar was seen, soft, uterus not palpable. P/V uterus was bulky, non-tender, mobile, adenexa free, os closed. 100gms of clots were removed from the vagina. The laboratory results were as follows: Hb-3.8gm%, PCV-14%, BT-1min 10 sec, CT- 3min, apTT-23 patient value and 31.6 control value, platelet count 2.8 lakh/mm3, Sr. creatinine-0.5mg/dl, RBS-189mg%, TC-18,500cells/mm3, DC-N87L11M2, ESR-55mm/hr, Sr.Na+-136mEq/L and Sr.K+-3.7mEq/L, Serum β Hcg:<2.39mIU/ml. The patient was immediately transfused with 3 units of A+ packed red cells. She was started on injection Cefotaxim and

injection metronidazole. After stabilizing the patient, imaging was done.

Trans-abdominal USG with colour Doppler showed well-defined rounded cystic lesion with colour uptake and turtulence measuring 1.3cmx1cm in anterior wall of lower uterine segment, uterus was 11cm×5cm (fig:1a). Spectral Doppler showed arterial wave form with biphasic pattern (fig:1b).

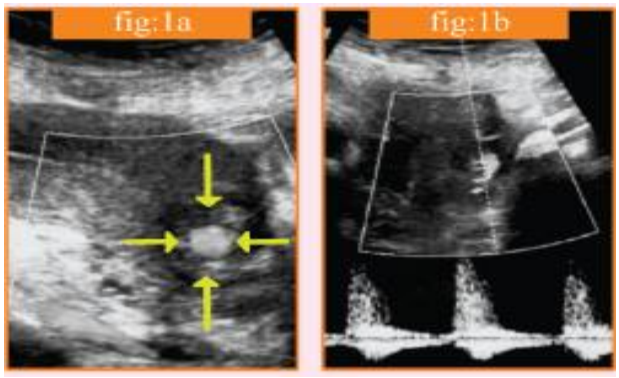

Fig 1: Trans-abdominal USG with colour Doppler

CT angiogram showed well circumscribed globular contrast filled lesion in left anterior myometrium with same attenuation as normal vascular enhancement in close relation to intramural branch of left uterine artery, suggestive of pseudoaneurysm of left uterine artery intramural branch. No active leak of contrast was seen (fig 2).

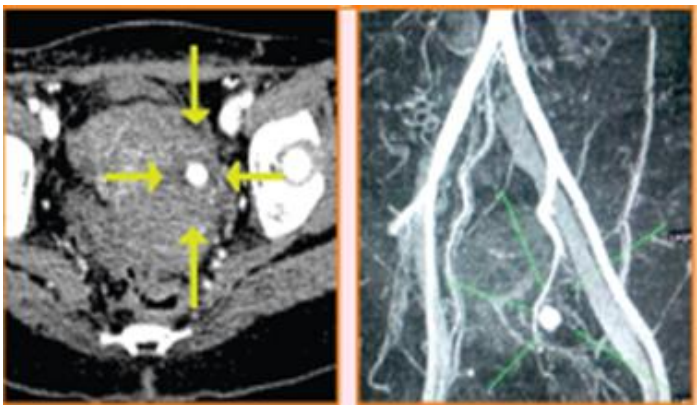

Fig 2: CT angiogram

Patient was referred to a centre with intervention radiology for uterine artery embolization. Selective transcatheter arterial embolization of pseudoaneurysm was planned and digital substraction angiography (DSA) was done. Left iliac arteriogram revealed a pseudoaneurysm from the terminal part of the left uterine artery (fig 3).

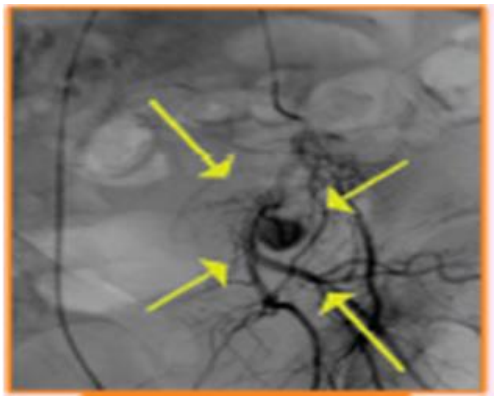

Fig 3: Terminal part of the left Uterine Artery

Superselection of terminal branch of left uterine artery was done using microcatheter and was embolized with N-butyl cyanoacrylate. The right uterine artery was embolized with gel foam. Post-operative period was uneventful and the bleeding stopped. Post embolization angiography showed no evidence of pseudoaneurysm (fig 4). Follow up USG with colour Doppler showed aneurysmal cavity filled with echogenic content with no evidence of blood flow.

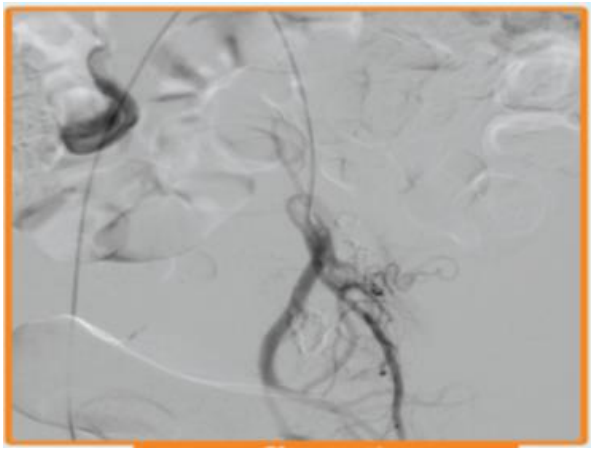

Fig 4: Post embolization angiography

Discussion

As reported by Registrar, General, India and Centre for Global Health Research; the most common direct causes of pregnancy-related mortality in India were hemorrhage (38%). Various studies in different regions of India report different prevalence of PPH. A study in North east region of India reported 94 maternal deaths/102525 live births, out of these, 53.19% women died due to hemorrhage accounting for about 21.27% of total deaths [8]. Similar studies reported prevalence of 22.7% in Delhi [9] and 28.57% in Orissa [10]. Hemorrhage was also found to be the major cause of maternal mortality in West India, accounting for 24.6% of maternal deaths in that region [11]. In Kerala it is 19.8%. Mukherji *et al* [12] reported that 58% of maternal deaths due to hemorrhage were actually due to PPH, resulting from lack of provision of emergency transport at community level. Though, PPH remains one of the major causes of maternal mortality.

A pseudoaneurysm forms due to surgical trauma: pelvic surgery, caesarean section or uterine curettage. Pseudoaneurysm results from an arterial wall laceration or a puncture that allows blood to enter periarterial tissue and create a perfused sac that communicates with the parent artery lumen. The pseudoaneurysms of the uterus that have been reported so far occurred at earlier postoperative stages [13]. Singh *et al* [14] reported a case in which a uterine pseudoaneurysm occurred at 40 days after cesarean section. In their case, the patient presented with history of excessive vaginal bleeding and haematuria since one day that required transarterial embolization. In our case, we also experienced an early occurrence of pseudoaneurysm; it took place 36th day after cesarean section with excessive bleeding per vagina for 1 day.

The initial diagnostic modalities for pseudoaneurysm are USG with color Doppler and CT angiography. We performed color Doppler ultrasound for the diagnosis of uterine pseudoaneurysm in our case because color Doppler ultrasound is useful in the detection of blood flow and its direction in a cystic structure.

Indeed, color Doppler ultrasound showed a high sensitivity and specificity in the detection of uterine pseudoaneurysms [13]. In our case, pseudoaneurysm was seen as anechoic lesion on USG and doppler demonstrates turbulent arterial flow, to-and-fro sign in the neck and yin yang sign (fig 5) in the body of pseudoaneurysm with a narrow neck.

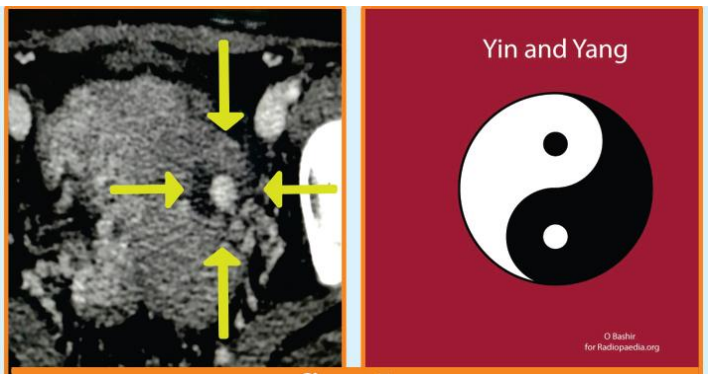

Fig 5: To-and-fro sign in the neck and yin yang sign

CT angiography is the gold standard for diagnosis, it can confirm the diagnosis and help rule out other more common causes of delayed postpartum hemorrhage. Also it may help in the design of definitive treatment strategies. In our case, CT angiogram revealed pseudoaneurysm of left uterine artery intramural branch.

Various treatment options are available for uterine pseudoaneurysm such as transarterial embolization of uterine artery (TAE), surgical ligation of uterine arteries, hysterectomy. TAE has been accepted as the safest choice for uterine pseudoaneurysm. Selective embolization of an affected vessel can be performed and bleeding can be arrested instantaneously. In 1979 Brown *et al*. [15] reported the first case of selective arterial embolisation used successfully to treat an extra uterine pelvic hematoma after 3 failed surgical attempts to control the bleeding. Since that time arterial embolisation has been used successfully to control postpartum bleeding due to atony, placenta accreta, extrauterine pregnancies and vulval and vaginal hematoma. TAE success rate is >90% due to its advantages of low morbidity, easy localization of bleeder, more distal occlusion than surgical ligation, decrease duration of hospital stay, no need of general anesthesia, repeat embolisation if needed can be performed and preservation of future fertility [14]. However, our experience suggests that urgent interventional therapy may not be necessary in all cases of uterine pseudoaneurysm. As more cases accumulate, a better understanding of its natural history will emerge, and more appropriate management will come to light.

Our patient developed a pseudoaneurysm at 36th day post-C/S delivery. Treatment was given by angiographic embolization, left uterine artery was embolized with N-butyl cyanoacrylate and right uterine artery was embolized with gel foam. In a retrospective review of eight women, Rosenthal and Colapinto [16] observed that angiographic arterial embolisation was the most useful clinical tool in the management of post-operative vaginal haemorrhage. Nanjundan *et al*. [6] have reported a similar case of secondary PPH on 19th post-operated day and were treated by embolization. Pelage *et al* [17] evaluated the efficacy and safety of selective arterial embolisation of uterine arteries in women with delayed secondary postpartum haemorrhage. In their study (N=14), pseudoaneurysms of the uterine artery were found in two participants and immediate resolution of external bleeding was observed after embolisation. In our study, there were no complications related to this invasive treatment. Other authors have described complications, including muscle pain and bladder necrosis [6].

Conclusion

Pseudoaneurysm of uterine artery should be considered as a differential diagnosis, when the patient presents with secondary PPH after caesarean section. Selective embolization of an affected vessel can be done to arrest the bleeding. Being a minimally invasive procedure, TAE should be offered whenever and wherever feasible. Although embolization is the treatment of choice, its use depends on the availability of interventional radiologists and angiographic equipment.

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