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Case Report

Unilateral Uterine Artery Embolization Therapy for Secondary Postpartum Hemorrhage in Pseudoaneurysma of The Sinistra Uterinary Artery Post-Cesarean Section

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KEYWORD
Uterine Artery Embolization; Postpartum Hemorrhage; Pseudoaneurysm

ABSTRACT

Introduction: Uterine artery pseudoaneurysm is one of the rare malformations of uterine blood vessels due to incomplete closure of the torn uterine artery wall. This condition accounts for about 3% of postpartum hemorrhage cases that could be life-threatening. Transcatheter arterial embolization has recently emerged as a safe and highly effective alternative treatment.

Case Presentation: A 30-year-old woman complained of abdominal pain accompanied by a large amount of blood from the birth canal, which was felt on the 28th day after a cesarean section. The patient underwent laparotomy and had grade III intra-abdominal adhesions. On CT-Angiography investigation, we found saccular type aneurysm in the left uterine artery, hematometra, and fluid collection on the right side of the periuterine, multiple paraaortic lymphadenopathies, hepatomegaly, and left adnexal cyst. With the pseudoaneurysm, the patient was planned to undergo uterine artery embolization. The patient was discarded. During follow-up, results of CT-scan pelvic angiography include dilated and convoluted right uterine artery.

Conclusion: In this case report, uterine artery embolization (UAE) was performed; this procedure was chosen because the effectiveness of embolization was 97% and could maintain the desire for future pregnancies. The fertility prognosis in this case report was quite good after unilateral uterine artery embolization was performed concerning regular menstrual cycles.

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INTRODUCTION

Secondary postpartum hemorrhage is defined as massive bleeding from the birth canal 24 hours after delivery up to 12 weeks [1]. Postpartum hemorrhage cases in developing countries reach 0.47% to 1.44% [2]. Various factors that cause postpartum hemorrhage include uterine atony/subinvolution, lower genital tract trauma, placental abnormalities undergoing conservative treatment, uterine abnormalities such as uterine myomas, blood clotting factor disorders, use of anticoagulant drugs, and abnormalities (malformations) in the uterine blood vessels [3].

Uterine artery pseudoaneurysm is one of the rare malformations of uterine blood vessels due to incomplete closure of the torn uterine artery wall. This condition accounts for about 3% of postpartum hemorrhage cases. The aneurysm sac usually has a division of a single layer of soft tissue supported by arterial blood pressure, and rupture is unpredictable [4]. Uterine artery pseudoaneurysms can occur after invasive surgeries, including cesarean sections, vaginal deliveries, myomectomy, hysterotomies, or dilatation

and curettage, which can cause vascular damage that can lead to pseudoaneurysms of the uterine artery or its branches (D&C) [5,6]. In contrast, other known causes of uterine artery pseudoaneurysms include hysterectomy and myomectomy. Vapors are rare but cannot be ruled out as a cause of delayed or secondary postpartum hemorrhage [5,6,7]. Massive hemorrhaging brought on by the rupture of a uterine artery pseudoaneurysm (UAP) can put a woman's reproductive health in peril and her life in danger, which requires an emergency laparotomy, unlike other conditions treated conservatively [7].

In patients with uterine artery, pseudoaneurysms can also show atypical clinical features, so emergency treatment cannot be maximized. When infected, the most common asymptomatic symptoms are vaginal bleeding, abdominal pain, hypovolemic shock, or fever. Such a broad set of symptoms can complicate the diagnosis of a pseudoaneurysm, especially in nontraumatic delivery or abortion. The broad spectrum of symptoms in uterine artery pseudoaneurysms should always be considered in the differential diagnosis of abnormal uterine bleeding. In this context, contrast-enhanced computed tomography (CT) should always be considered in the diagnostic workup, as it plays a crucial role in determining the indications for digital subtraction angiography (DSA) [8]. Traditional surgical management of patients with uterine artery pseudoaneurysms includes revision procedures with packing, bilateral internal iliac ligation, or uterine artery ligation. If other treatments fail, then the final treatment is hysterectomy. Transcatheter arterial embolization has recently emerged as a safe and highly effective alternative treatment [7]. Uterine artery pseudoaneurysms are an underreported cause of vaginal bleeding, and the incidence of bleeding emergencies due to uterine artery pseudoaneurysms is unknown. This condition will cause vaginal bleeding that can be life-threatening, so proper diagnosis and treatment are needed. Thus, this article aimed to present a case of unilateral uterine artery embolization therapy for secondary postpartum hemorrhage in pseudoaneurysm of the sinistra uterinary artery post-cesarean section.

CASE PRESENTATION

A 30-year-old woman presented a post-SC day- 28 referral diagnosis with secondary Hemorrhagic Post-Partum e.c. susp. Endometritis + post laparotomy day-1 with intra-abdominal adhesions (failed hysterectomy). Previously, the patient underwent Section Caesarea (SC) surgery with indications of a former SC and was treated for four days without any additional complaints at the referral hospital. After 20 days, the patient complained of abdominal pain accompanied by a large amount of blood from the birth canal. The procedure was carried out by giving four flasks of PRC transfusion and three-days hospitalization. Nineteen days later, the patient complained of the same symptoms and received a transfusion of 3 flasks. After two days post-transfusion, the patient complained of re-bleeding from the birth canal as much as two under pads; the patient was planned for surgery to hysterectomy. Grade III intra-abdominal adhesions were found during the operation, so the hysterectomy failed. The patient finally received treatment at the Saiful Anwar hospital and improved when she left the hospital.

Complaints reoccurred after one-month post-hospitalization. The patient complained of the same thing again, namely bleeding from the birth canal as much as three under pads, and was diagnosed with AUB e.c endometritis + recurrent Fluxus + anemia (improvement). During hospitalization at the referring hospital, the patient received therapy with ceftriaxone injection of 2x1 gram, metronidazole injection of 3x500mg, tranexamic acid injection of 3x500mg, and oral misoprostol 3x1 tablets, and Packed Red Cells (PRC) transfusion of 8 flasks. Due to recurrent vaginal bleeding and the patient's clinical deterioration (blood pressure 70/palpation mmHg, weak pulse 110 beats/minute), the patient was referred to Saiful Anwar Hospital. The patient underwent CT angiography of the pelvis and found a saccular type aneurysm in the left uterine artery, hematometra, and fluid collection on the right side of the periuterine, multiple paraaortic lymphadenopathies, hepatomegaly and left adnexal cyst. The patient was planned for left uterine artery embolus. After the action, the complaints are reduced to allow the patient to control the clinical polity. During clinical follow-up, the patient underwent CT-scan pelvic angiography, and the results of dilatation and tortuous of the right uterine artery were found. The patient received an elective hysteroscopy in the third hospitalization with good results.

It was known that the patient had no history of hypertension, history of diabetes mellitus, history of asthma, or heart disease. History of defecating within normal limits and urinating was within normal limits. History of fever, cough, runny nose and contact with COVID-19 patients was denied. History of connection with confirmed/probable COVID-19 case refuted. History of previous delivery, the first child was born at term; baby weight was reached at 3,000 grams with a cesarean section for indication of prolonged 1st stage; the child is currently six years old. The second child was born at term gestational age, the baby's weight was taken at 3,000 grams with an SC action on the indication of a former SC, and the current age is 1.5 months.

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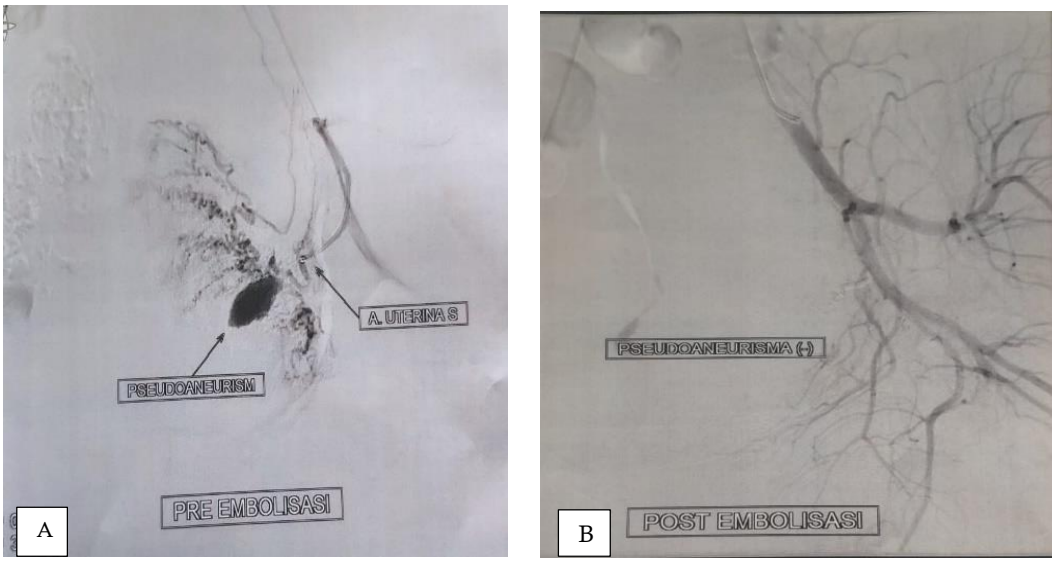

Fig. 1. A. Pre-embolization; B. Post-embolization

Investigation

First Hospitalization

On the first physical examination, the patient appeared to be moderately ill with compos mentis consciousness. The patient's height is 160 cm, and her weight is 65 kg. Blood pressure was also 130/91 mmHg, pulse 103x/minute, breathing 20x/minute, and axillary temperature 36ºC. Abdominal examination revealed a surgical wound covered with dry gauze, soft uterine fundus height central symphysis, good uterine contractions, midline scar (+), *and Pfannenstiel* scar (+). Examination of the external genitalia revealed Fluxus; it obtained minimal flux with closed and slippery portions when inspected. A vaginal touch examination revealed the Fluxus and size of the uterine cavity according to the gestational age of 12-14 weeks. On laboratory examination, the patient's Hb level was 10.4 g/dl, and leukocytes were 14,650 x103/µL. Abdominal ultrasound revealed fluid collections in bilateral periuterine and Douglas cavities.

Second Hospitalization

In the second hospitalization, the patient was moderately ill with compos mentis consciousness, blood pressure 105/74 mmHg, pulse 92x/minute, respiratory rate 20x/minute, oxygen saturation 99% with nasal cannula three-liter per minute. The results of physical examination on the abdomen were palpable cystic mass size 5x7 cm, flat surface, no clear boundaries, limited mobility, and scar pfannenstiel (+). Laboratory examination showed a hemoglobin level of 10.5 g/dl. Abdominal ultrasound revealed a left adnexal complex cyst and minimal ascites. On the eighth day of treatment, the patient underwent a CT-angiography examination. The results were obtained saccular type aneurysm in the left uterine artery, hematometra, fluid collection on the right side of the periuterine, multiple paraaortic lymphadenopathies, hepatomegaly, and left adnexal cyst. The patient was planned for left uterine artery embolization.

Third Hospitalization

In the third patient condition, the patient was moderately ill, compos mentis consciousness, with blood pressure 120/88mmHg, pulse 87x/minute, respiration 20x/minute, and axillary temperature 36C. On examination, the abdomen differs from the aforementioned hospitalized conditions. Currently, the uterine fundal height is difficult to evaluate and does not lump. On laboratory examination, it showed an improvement in hemoglobin to 12.4 g/dl. CT scan angiography showed dilatation and tortoise in the right uterine adnexa and its branches; the left uterine artery aneurysm was no longer visible. There was hemometry and a left adnexal cyst.

Treatment

In the first hospitalization, the patient was admitted with a diagnosis of P2002Ab000 PP SCTP on day 47 for indication of former SC + late hemorrhagic postpartum (HPP) e.c endometritis + post laparotomy on day 1 for representation of late HPP with intra-abdominal adhesions grade III (open and close), the patient received 20 IU oxytocin drip therapy in 500 cc RL (20 drops per minute for 24 hours), ceftriaxone injection 2x1 gram, metronidazole injection 3x500 gram, tranexamic acid injection 3x500mg and metergin injection 3x1 ampoule. The patient had experienced improvement, but the same complaint occurred one month later.

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Due to the bleeding did not stop, both patients were hospitalized with tampons and an evaluation 24 hours after tampon placement. In addition, the patient was also planned for an elective hysteroscopy. In this second hospitalization, the patient was also subjected to CT - angiography examination and found to have an aneurysm. The patient was continued with a hysteroscopy evaluation as a continuation of the previous procedure in the third hospitalization.

Outcomes and Follow Up

The results of an elective hysteroscopy at the last hospitalization showed that the cervix and vagina were in good condition, no lesions were found, the uterus was in an anteflexion position, and the intact endometrium had no lesions. It was smooth, and there were no abnormalities in the upper uterine segment to the lower uterine segment; ostium right and left tubes are visible and function types, and there are no varicose veins on the endometrial picture.

DISCUSSION

In this case, the patient has been hospitalized twice at Saiful Anwar Hospital with varying diagnoses. At the beginning of the first hospitalization, this patient was diagnosed with Late HPP e.c endometritis + Post laparotomy on day 1 to indicate late HPP with intra-abdominal adhesions gr.III (open and close) + P2002Ab000 postpartum section day 47 showed a former cesarean section. Meanwhile, at the beginning of hospitalization, both patients were diagnosed with Abnormal uterine bleeding- non-classified (AUB-N) + recurrent Fluxus + Cystoma ovary + Former section twice without a known cause of AUB-N, until the final diagnosis with AUB-N due to Pseudoaneurysm A. uterine Sinistra + Cystoma ovary + former cesarean section.

The long process of establishing the diagnosis in this patient was because of the absence of a pelvic angiography CT-Scan from the patient's initial arrival. The patient only underwent an abdominal ultrasound examination due to a suspicion of late HPP with endometritis. A laparotomy was performed to find causes of bleeding other than endometritis. Regarding the diagnosis of the first hospitalization with late HPP due to endometritis and the second hospitalization with AUB-N, whose cause was not established, this patient was given therapy that did not stop bleeding treatment based on the management of acute bleeding and was only given anti-fibrinolytic treatment. After the patient experienced repeated hypovolemic shock at the referring hospital and abdominal ultrasound revealed no significant abnormalities, the diagnosis of AUB-N caused by the left uterine artery pseudoaneurysm could be made.

Regarding the diagnosis of uterine artery pseudoaneurysm, Chaudhry reported that uterine artery aneurysm should be one of the differential diagnoses that can be considered in pregnant women who present with vague bladder symptoms or radiating pelvic pain [9]. Several authors have reported that patients with uterine artery pseudomembranes usually present with vaginal bleeding, and the diagnosis of pseudomembranes can be confirmed by color doppler ultrasound examination [10]. A pseudoaneurysm will appear as a saccular artery pocket or an abnormally dilated artery with a surrounding hematoma [11]. Angiography is a valuable imaging method for confirming the diagnosis of uterine artery pseudoaneurysm. An angiographic procedure will be performed to confirm the diagnosis based on the color Doppler ultrasound examination results, indicating a suspected uterine artery pseudoaneurysm. The angiographic examination was performed by inserting a catheter through the right femoral artery, and the angiogram image showed focal dilatation of the right uterine artery. This finding represents the characteristic feature of uterine artery pseudoaneurysm [12]. In this case report, the diagnosis of uterine artery pseudomembrane is based on clinical manifestations in the form of repeated vaginal bleeding after cesarean section, which causes recurrent hypovolemic shock and failure of medical therapy using anti-fibrinolytic and inadequate operative therapy. The patient had undergone CT-scan angiography and had findings in the form of a saccular aneurysm in the left uterine artery with dome width + 7.6 mm, height + 11 mm, neck width + 1.5 mm, which supported the diagnosis of AUB-N—caused by the pseudomembrane of the left uterine artery.

Uterine artery pseudoaneurysms are usually the result of traumatic injury to the uterine artery wall. Inflammatory lesions and arterial infections might result from a variety of surgeries. A localized hematoma and pseudoaneurysm might develop as a result of improper suturing procedures damaging the artery that supplies blood to the uterine myometrial region. Damage can result in the shearing of the artery walls, allowing arterial blood to build up between the vessel wall layers If this blood accumulation continues to communicate with the arteries and is surrounded by the tunica adventitia (the outermost layer of the vessel wall), a pseudoaneurysm is formed. Meanwhile, aneurysms have a three-layered fence, which can distinguish them from pseudoaneurysms [4]. Extraluminal turbulent flow within a pseudoaneurysm can cause enlargement and possibly even rupture of the pseudoaneurysm if not recognized [8].

In some cases, uterine artery pseudoaneurysms are also misunderstood as remnants of conception or changes in routine menstruation during pregnancy, infection, or trauma to the endometrium. Acquired

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uterine artery pseudoaneurysms may be a biological aberration of placental bed regression at placentation [13]. Meanwhile, postoperative uterine artery pseudoaneurysms may be caused by a mixed or poorly attached endometrium that triggers a local immune response and activates inflammatory cytokines that lead to endometrial-myometrial biologic changes [14]. In this case report, the possible etiology is iatrogenic due to trauma in the form of cesarean section, but it is likely due to biological deviation from placental bed regression.

The initial management of a ruptured uterine artery pseudoaneurysm is similar to other acute blood loss cases. The main principle is to obtain intravenous access and resuscitation using fluids and blood products. Insertion of vaginal tampons or insertion of intrauterine tampons until definitive treatment can be carried out [15]. Treatment of uterine artery pseudoaneurysms is tailored to the clinical presentation, age, and desire to maintain future fertility. Not all patients with uterine artery pseudoaneurysms undergo surgery in the form of hysterectomy or internal iliac artery ligation, but embolization can be considered in patients aiming for conservative treatment [16,17]. The treatment for uterine artery pseudoaneurysm is the uterine artery embolization procedure because it is safe and effective. With the development of science in interventional radiology, the "Transcatheter Uterine Artery Embolization" procedure is a minimally invasive procedure that allows conservative management in cases of secondary postpartum hemorrhage [5,18]. The success rate of uterine artery embolization in cessation of secondary postpartum hemorrhage is 97%, with several advantages such as accurately identifiable bleeding sites, repeat embolization if necessary, and embolization will not affect bleeding control measures to avoid the risks of general anesthesia. Embolization will be performed on arteries with unilateral uterine artery pseudoaneurysms, but if bleeding persists, bilateral embolization can be achieved [15,19].

Embolizing agents can be divided into temporary agents and permanent agents. The choice of an embolizing agent depends on the embolization site, the intended purpose, the type of vessel, the characteristics of the blood flow, and the potential for complications [20]. NBCA has emerged as a potent permanent embolic agent in treating acute bleeding of various etiologies due to its rapid intravascular polymerization [21]. NBCA as a 'glue' agent in the embolizing procedure, enters blood arteries, interacts with cations there, and polymerizes to form a solid material that occludes the vascular lumen and achieves hemostasis is the mechanism of NBCA embolization [22]. One study found that NBCA was effective in patients with postpartum hemorrhage due to coagulopathic disorders, pseudoaneurysms, or extravasation. The use of permanent NBCAs can cause concerns about patient fertility, but the success rate in cases of postpartum hemorrhage due to pseudoaneurysms or aneurysms reaches 67.9%-92.3% [23]. The choice of uterine artery embolization therapy in cases of uterine artery pseudoaneurysm depends on various factors such as the degree of bleeding, the shape of the pseudoaneurysm, the size of the pseudoaneurysm, future fertility, and complications such as thromboembolism and vesicovaginal fistula. If embolization is performed unilaterally, evaluation using angiography for the contralateral side is necessary [24]. Complications of embolism using glue in the management of pseudoaneurysms include infection due to the insertion of a foreign body during the procedure, causing acute inflammation to fibrosis in various places, embolization from non-target organs due to improper catheter positioning, reflux of embolic agents, and during embolization, which can result in rupture of the pseudoaneurysm. After embolization using glue will increase the risk of temporary or permanent amenorrhea, abortion, and even infertility due to hypoperfusion into the uterus [25]. The patient was evaluated six months after embolization without any signs and symptoms of embolization in the lower target organs or menstrual disorders (amenorrhea).

Vilos et al. stated that the pregnancy success rate after uterine artery embolization was lower than in patients undergoing myomectomy for uterine myomas, thus recommending embolization for women who do not wish to have a future pregnancy and uterine artery embolization as a second-line option. In contrast to the Society of Interventional Radiology (SIR), which states that the desire for pregnancy is not a relative contraindication to uterine artery embolization (UAE) [26]. This case report found that the menstrual cycle returned to normal to maintain a successful pregnancy [27]. However, it can be considered that a successful pregnancy in this patient still has the risk of massive bleeding, preterm labor, and postpartum hemorrhage because the CT-scan angiography examination revealed dilatation and tortuous of the contralateral uterine artery.

CONCLUSION

In this case report, the diagnosis of uterine artery pseudoaneurysm can be screened through color Doppler ultrasound and CT scan angiography as confirmation. In this case report, the etiology of uterine artery pseudoaneurysm is iatrogenic, which can be caused by cesarean section or biological deviation from placental bed regression. In this case report, uterine artery embolization (UAE) was performed. This procedure was chosen because the effectiveness of embolization was 97% and could maintain the desire for future pregnancies. The choice of glue as an embolization

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agent is suitable for use in this case report of uterine artery pseudoaneurysm because the glue is permanent occlusive and can be used in conditions of tortuous target blood vessels. In this case report, there was no recanalization of the embolized arteries due to an embolic agent (glue), which can result in permanent occlusion. Due to fertility considerations, in this case report, unilateral arterial embolization was performed. At the same time, the results of the CT-scan angiography evaluation five months after the procedure revealed dilatation and tortuous of the contralateral uterine artery, so the patient is currently being treated conservatively based on complaints physical examination and investigations. Periodically. If a pseudoaneurysm is found in the contralateral uterine artery, contralateral uterine artery embolization can be performed. The fertility prognosis in this case report was quite good after unilateral uterine artery embolization was performed regarding regular menstrual cycles. Although the prognosis for fertility is quite good, the risk of bleeding during pregnancy, preterm labor, and massive postpartum hemorrhage can be considered.

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CONFLICT OF INTEREST

The writers claim to be free of any conflicts of interest.

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