



Case report

Pelvic hematoma: The same complication with different etiology after patent foramen ovale closure

Guohui Liu^{a,b}, Chang Wang^{a,b}, Hongliang Yang^{a,b}, Ming Yu^{a,b,*},¹, Ping Yang^{a,b,**},¹

^a Department of Cardiology, China-Japan Union Hospital of Jilin University, China

^b Jilin Provincial Cardiovascular Research Institute, Jilin Provincial Engineering Laboratory for Endothelial Function and Genetic Diagnosis of Cardiovascular Disease, Changchun, Jilin Province, 130031, China

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ABSTRACT

Well-known complication associated with patent foramen ovale (PFO) closure include infection, acute cardiac tamponade, and local complications such as adjacent arterial or nerve damage, hemorrhage, and thrombophlebitis. Pelvic hematoma is rare and potentially fatal complication. This paper reports two cases of severe hemorrhagic shock within 1 day after PFO closure. Both female patients presented to our department with history of headaches and were diagnosed with PFO. Both patients underwent percutaneous PFO closure from the right femoral vein. One day after the procedure, both patients experienced pelvic hematoma and were successfully rescued by compression hemostasis and uterine artery embolization. Both patients recovered well during follow-up. Life-threatening pelvic hematoma associated with PFO closure has a certain incidence and should be considered. Peripheral vascular complications after PFO closure can be safely treated but should not be ignored. We believe that the prevention of vascular mechanical damage during surgery is important. The possibility of spontaneous uterine artery rupture should be considered for unexplained pelvic hematoma. Although it is a rare complication, severe bleeding after PFO closure remains unpredictable. Timely and correct diagnosis and appropriate treatment are required. If the timing is delayed, there could be serious consequences.

1. Introduction

Patent foramen ovale (PFO), an embryonic remnant of fetal circulation, is present in 20–25 % of adults [1]. Although frequently clinically asymptomatic, PFO can play a pathological role by allowing thrombus transfer from the veins to the systemic circulation (paradoxical embolism). The incidence of PFO in migraine patients is 30–40 %, and in migraine patients with aura, it is 48–70 %. Recent clinical studies have described the efficacy and safety of percutaneous PFO closure for treating migraine with aura [2] and secondary prevention of cryptogenic stroke [3]. Complications of PFO closure include ectopic embolism, acute cardiac tamponade, atrioventricular regurgitation, infective endocarditis, and vascular complications, such as arterial injury, venous thrombosis, arteriovenous fistula, and hemorrhage. Few studies have reported the occurrence of pelvic hematomas after PFO closure. Although this complication often occurs after arterial puncture above or below the inguinal ligament, there are few reports due to femoral venous

* Corresponding author. Department of Cardiology, China-Japan Union Hospital of Jilin University, Changchun, 130033, China.

** Corresponding author. Department of Cardiology, China-Japan Union Hospital of Jilin University, Changchun, 130033, China.

E-mail addresses: yuming2019@jlu.edu.cn (M. Yu), pyang@jlu.edu.cn (P. Yang).

¹ Ming Yu and Ping Yang contributed equally to this work.

puncture. Pelvic hematoma due to spontaneous uterine artery rupture is less frequently reported. Herein, we report two cases of pelvic hematoma after patent foramen ovale closure. The causes of hematoma formation and possible ways of hematoma diffusion were discussed, and suggestions for avoiding such complications were put forward.

1.1. Case 1

A 29-year-old Chinese woman presented to our department with a 10-year history of headaches and aggravation of fatigue for 1 month. The patient did not report any relevant medical history. On admission, a physical examination revealed a blood pressure of 101/70 mmHg and a heart rate of 84 beats per minute (bpm). Initial blood examinations showed a hemoglobin level of 137 g/L, hematocrit of 40.5 %, prothrombin time (PT) of 11 seconds, and activated plasma thromboplastin time (APTT) of 34.6 s. Other blood tests revealed no significant abnormalities. Cranial magnetic resonance imaging (MRI) did not reveal any intracranial pathology. Contrast transthoracic echocardiography demonstrated a patent foramen ovale (PFO) with a large right-to-left shunt. The patient underwent percutaneous PFO closure from the right femoral vein for secondary prevention of cerebrovascular events. A 24 mm MemoPart™ PFO occluder (Lepu, Beijing, China) was deployed under the transthoracic echocardiography (TTE) guidance. Heparin (5000 IU) was used during the operation, and low-molecular-weight heparin, aspirin, and clopidogrel hydrogen were administered postoperatively.

Four hours after the procedure, the patient complained of pain in the right lower abdomen, associated with severe hypotension and tachycardia. Her hemoglobin level dropped to 76 g/L, and her hematocrit decreased to 22.5 %, which was treated with a blood transfusion and intravenous norepinephrine drip. Vascular ultrasonography of both lower limbs showed that the femoral artery, femoral vein, iliac artery, and iliac vein had smooth blood flow with no dissection, arteriovenous fistula, or pseudoaneurysm. There was effusion between the muscular layer and the peritoneum of the right abdominal wall (7.0×0.6 cm), and there was a hematoma behind the muscular layer of the pelvic abdominal wall ($12.8 \times 8.3 \times 10.7$ cm) (Fig. 1A–B). Vascular contrast-enhanced ultrasonography revealed a heterogeneous echogenic mass adjacent to the right iliac vessel (10.7×6.8 cm) (Fig. 1C). The origin of the active hemorrhage was suspected to be a branch of the right external iliac vein. Abdominal computed tomography (CT) revealed a mixed-density hematoma in the right colonic sulcus and pelvic cavity (Fig. 1D). The hematoma with a locally detectable blood flow signal was detected in ultrasonography (Fig. 2A). Low-molecular-weight heparin, aspirin, and clopidogrel hydrogen were discontinued immediately, and hemorrhage was stopped with right lower abdominal compression under ultrasound guidance for 2 days. The patient was managed conservatively with a blood transfusion and did not require endovascular repair because of her stable vital signs. Follow-up vascular ultrasound showed a significant decrease in hematoma behind the muscular layer of the pelvic abdominal wall (8.0×9.0 cm) and no blood flow signal in the hematoma (Fig. 2B).

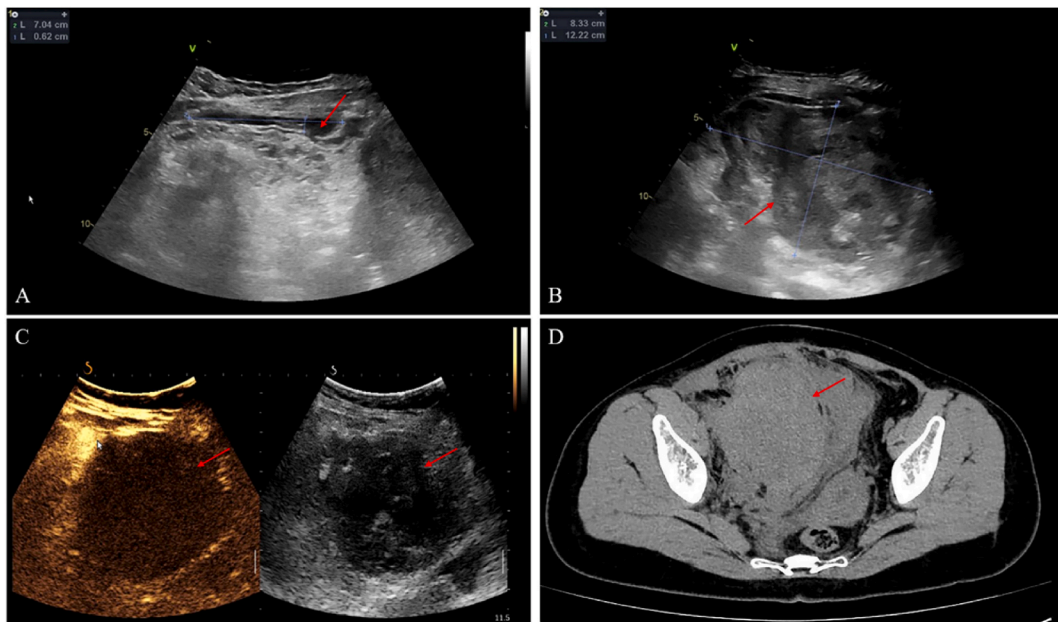


Fig. 1. Pelvic hematoma of case 1. A. Effusion between the muscular layer and the peritoneum of the right abdominal wall (7.0×0.6 cm) (red arrow). B. Hematoma behind the muscular layer of the pelvic abdominal wall ($12.8 \times 8.3 \times 10.7$ cm) (red arrow). C. Vascular contrast-enhanced ultrasonography revealed a heterogeneous echogenic mass adjacent to the right iliac vessel (10.7×6.8 cm) (red arrow). D. Abdominal CT revealed a hematoma in the pelvic cavity (red arrow).

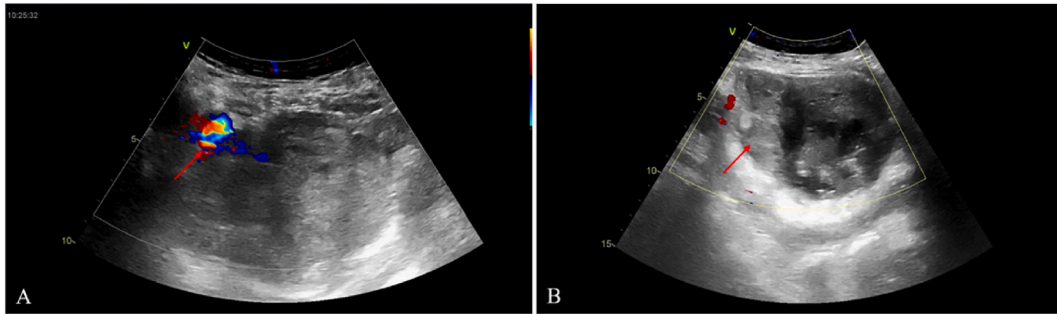


Fig. 2. Changes in blood flow of hematoma in case 1. A. A locally detectable blood flow signal in hematoma one day after procedure (red arrow). B. A significant decrease in hematoma (8.0 × 7.0 × 9.0 cm) and no blood flow signal in the hematoma during follow-up (red arrow).

1.2. Case 2

A 32-year-old woman with no underlying diseases presented to our department after 20 years of intermittent temporal and occipital headaches. Her blood pressure was 110/81 mmHg, and her heart rate was 82 bpm. The initial blood examinations showed a hemoglobin level of 107 g/L, hematocrit of 32.1 %, PT of 12.2 s, and APTT of 23.2 s. Other blood tests revealed no significant abnormalities. Cranial MRI revealed bilateral central semioval and frontal lobe infarcts (Fig. 3A). Transcranial Doppler with intravenous injection of agitated saline microbubbles demonstrated a large right-to-left shunt (Fig. 3B). Contrast transthoracic echocardiography demonstrated a PFO with a large right-to-left shunt (Fig. 4A). She underwent percutaneous PFO closure from the right femoral vein to prevent the recurrence of cerebrovascular events. A 25 mm Amplatzer PFO occluder (Abbott, Chicago, IL, USA) was deployed under transthoracic echocardiography (TTE) guidance. Heparin 5000 IU was used intraoperatively, and low-molecular-weight heparin, aspirin, and clopidogrel hydrogen were administered postoperatively.

Three hours after procedure, there was tenderness in the hypogastric region and a decrease in systolic blood pressure to 80 mmHg. Hemoglobin dropped to 59 g/L, and hematocrit decreased to 17.6 %, which was treated with blood transfusion and fluid infusion. Abdominal color ultrasonography showed a hematoma behind the muscle layer of the pelvic abdominal wall (6.42 × 8.65 cm) (Fig. 4B). Abdominal CT revealed a hyperdense hematoma on the right side of the pelvic cavity (Fig. 4C), and contrast-enhanced CT confirmed pelvic hematoma and effusion (Fig. 4D). The origin of the active hemorrhage was suspected to be a branch of the right internal iliac artery. Low-molecular-weight heparin, aspirin, and clopidogrel were immediately discontinued. Urgent internal iliac arteriography through the left femoral artery showed leakage of iodinated contrast agent from the right uterine artery (Fig. 5A) and obvious tortuosity in the right ovarian artery (Fig. 5B). The patient underwent uterine and ovarian artery embolization with absorbable gelatin sponge granules. Post-embolization angiography revealed no active contrast agent extravasation (Fig. 5C–D). The patient was discharged in stable general condition without further incident, and her hemoglobin level returned to 99 g/L. Contrast-enhanced abdominal CT over 1 week showed that the hematoma was smaller than before, the vascular shadow in the hematoma disappeared, and intraperitoneal and retroperitoneal effusions were significantly reduced.

2. Discussion

Numerous observational studies have demonstrated an association between PFO and ischemic stroke. PFO closure as a therapeutic modality is an exciting area of research. The role of PFO closure in preventing stroke and recurrence is well-established. PFO closure is effective in migraine patients who do not respond to conventional medical therapy [4].

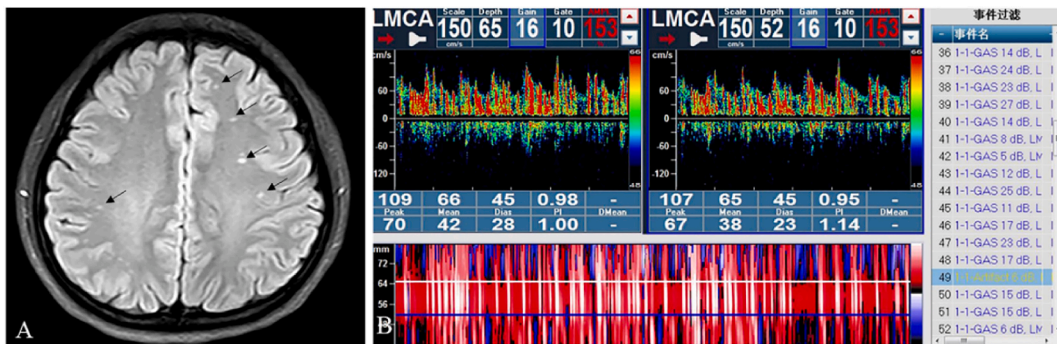


Fig. 3. Cranial examinations of case 2. A. Cranial MRI revealed bilateral central semioval and frontal lobe infarcts. B. Transcranial Doppler with intravenous injection of agitated saline microbubbles demonstrated a large right-to-left shunt. MRI: magnetic resonance imaging.

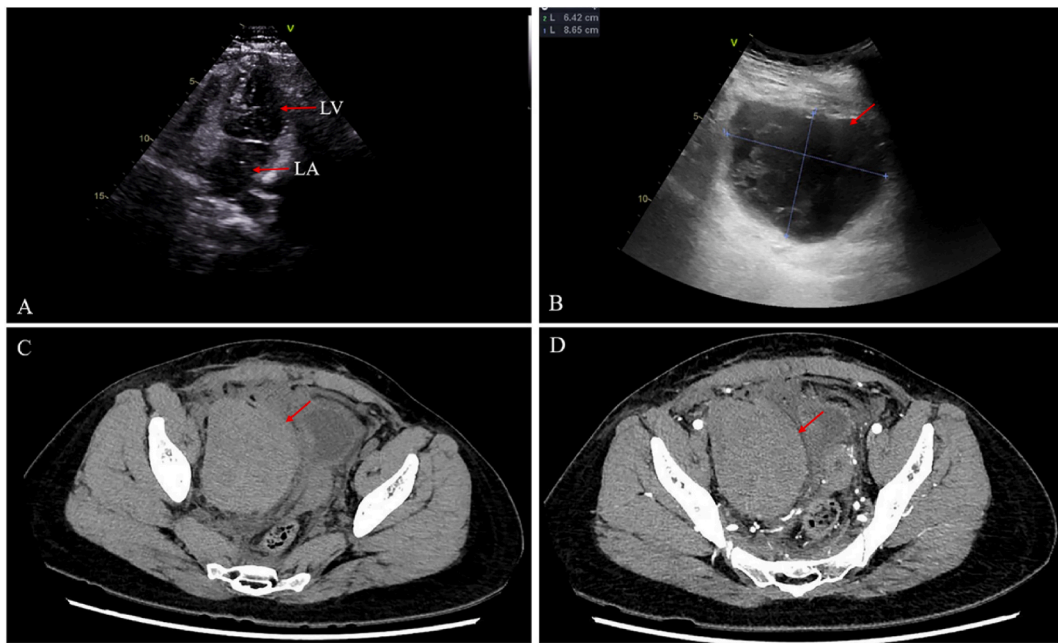


Fig. 4. Echocardiography and pelvic hematoma of case 2. A. Contrast transthoracic echocardiography demonstrated a PFO with a large right-to-left shunt. B. Abdominal color ultrasonography showed a hematoma behind the muscle layer of the pelvic abdominal wall (6.42 × 8.65 cm) (red arrow). C. Abdominal CT revealed a hyperdense hematoma on the right side of the pelvic cavity (red arrow). D. Contrast-enhanced CT confirmed pelvic hematoma and effusion (red arrow). LA: left atrium; LV: left ventricle; CT: computed tomography.

Femoral venous puncture is the most common procedure used for percutaneous PFO closure. This procedure is usually performed blindly by manually locating the femoral pulse according to the external markers. Mechanical complications associated with this technique include arteriovenous fistulas, pseudoaneurysms, vascular perforation with severe bleeding, and nervous system damage. It has been reported that major hematoma formation as a mechanical complication of femoral vein cannulation occurred in only 1.3 % of cases, the predominant cause being adjacent artery puncture [5]. There are few reports of pelvic cavity hematomas caused purely by venous puncture. In case 1, the authors identified that the formation of a pelvic cavity hematoma was a complication of femoral venous cannulation. This complication may be attributed to a high superior inguinal double-wall puncture of the femoral vein or an incidental injury of iliac vein's branch. Vascular ultrasound showed no injury to the femoral artery, which would have caused such a large pelvic hematoma. The blood transmission path of the femoral venous puncture can be explained according to the anatomy of the femoral sheath and the triangle. Anteriorly, the sheath wall is formed by the continuation of the transverse fascia and is directly connected to the anterior abdominal wall. Posteriorly, it is formed by the iliac fascia and directly communicates with the retroperitoneum. Bleeding in the femoral sheath follows the path of least resistance up into the prevesical space. It spreads anteriorly along the apical peritoneum beneath the transverse fascia to the anterior abdominal wall [6]. In this case, the needle puncture was a double-walled puncture above the inguinal ligament, and postoperative bleeding was most likely to pass through the posterior wall of the external iliac vein. Heparinization during PFO closure aggravates bleeding. The hematoma and bleeding site was found in time, and manual compression could stop the bleeding, avoiding surgical intervention.

Although pelvic cavity hematoma is usually associated with trauma or iatrogenic injury, spontaneous pelvic cavity hematoma is rare and may occur because of vasculopathy, coagulopathy, or pathology. Spontaneous rupture of the uterine artery is an extremely rare and potentially life-threatening cause of pelvic cavity hemorrhage. Case reports have described spontaneous pelvic cavity hemorrhage secondary to uterine varicose veins or utero-ovarian vessel ruptures. The most common location of vein rupture was the broad ligament (78.3 %), followed by the dorsal uterus (18.3 %) and the anterior uterus (3.3 %) [7]. However, similar data were not available for arterial ruptures. The most common causes of arterial rupture are the uterine artery or pseudoaneurysms. In case 2, the patient presented with a sudden decrease in blood pressure and an increase in heart rate 3 hours after PFO closure. An emergency enhanced CT scan revealed active extravasation of contrast material in a hyperdense mass in the pelvis, consistent with spontaneous rupture of the uterine artery, although it was not specific. Subsequently, angiography was performed to confirm the diagnosis and treat the ruptured uterine artery. We identified three case reports in PubMed, including pelvic cavity hemorrhage due to spontaneous rupture of uterine arteries in non-pregnant and puerperium women [8–10]. To the best of our knowledge, this is the first reported case of spontaneous uterine artery rupture after PFO closure which was induced by heparin. Digital subtraction angiography (DSA) is the gold standard for diagnosing spontaneous uterine artery rupture. When the patient has active bleeding, contrast extravasation can be observed, which can directly guide embolization. Transcatheter uterine artery embolization is the preferred treatment for uterine artery malformations after pelvic angiography [11]. It is a safe treatment with a high success rate and few reported complications [12], which can prevent hysterectomy.

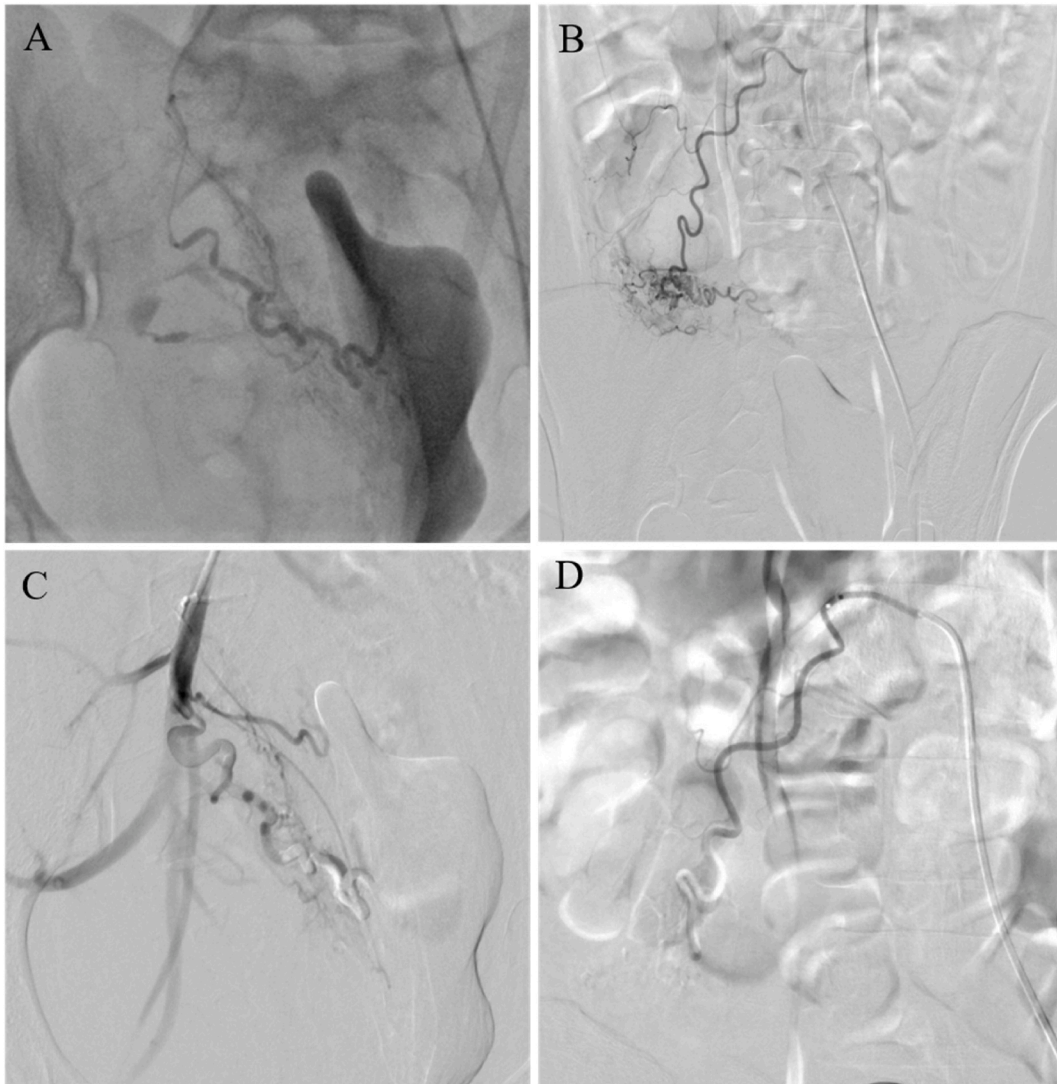


Fig. 5. Internal iliac arteriography of case 2. A. Leakage of iodinated contrast agent from the right uterine artery. B. Obvious tortuosity in the right ovarian artery. C. Post-embolization angiography of uterine artery. D. Post-embolization angiography of ovarian artery.

Pelvic cavity hemorrhage after PFO closure is extremely rare. The immediate detection of complications is essential to prevent life-threatening bleeding. Most fatal cases are associated with inadvertent injury, delayed diagnosis, or incomplete diagnosis of vascular lesions. Predisposing factors for hemorrhagic risk include underlying disease, anatomical variations, and misplacement of catheterization. In addition, using heparin during surgery and antiplatelet therapy after surgery should also be considered. The most common symptoms were hypotension, bellyache, and reduced hemoglobin and hematocrit levels. Treatment consists mainly of managing the acute complication of the bleeding (hypovolemia or shock), identifying the cause as soon as possible, and treating the primary cause of pelvic cavity hemorrhage. The femoral venous puncture should be below the level of the inguinal ligament, where the vessel can be compressed for hemostasis, and the femoral head should be searched during palpation before the puncture. Imaging guidance can be sought if necessary. Understanding the spread of hematoma and its timely identification is critical for proper management. Moreover, the possibility of spontaneous uterine artery rupture should be considered for unexplained pelvic hemorrhage. Although it is a rare complication, severe bleeding after PFO closure remains unpredictable. Timely and correct diagnosis and appropriate treatment are required. If the timing is delayed, there could be serious consequences.

Ethics statement

The studies involving human participants were reviewed and approved by the China-Japan Union Hospital of Jilin University Ethics Committee. The ethics committee waived the requirement of written informed consent for participation.

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Data availability statement

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding authors.

CRediT authorship contribution statement

Guohui Liu: Conceptualization, Data curation. **Chang Wang:** Data curation, Resources. **Hongliang Yang:** Resources. **Ming Yu:** Conceptualization, Data curation, Writing – original draft, Writing – review & editing. **Ping Yang:** Supervision.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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