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Case Report

A case of a patient who underwent transcatheter arterial embolization for unruptured splenic aneurysm during pregnancy ☆,☆☆

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ABSTRACT

Here, we report the case of a 30-year-old female patient who underwent coil embolization for unruptured splenic artery aneurysm without any complication at 26 weeks of pregnancy with reduction in fetal radiation exposure. The patient did not suffer from rupture of splenic artery aneurysm during or after procedure. Pregnancy is a risk factor of splenic artery aneurysm rupture with a high mortality rate. Transcatheter arterial embolization at 26 weeks of pregnancy might be a better treatment alternative for a pregnant patient with splenic artery aneurysm with respect to the endurance of fetal radiation exposure to prevent aneurysm rupture.

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Introduction

Splenic artery aneurysms accounts for 4%-5% of systemic aneurysms and 60% of visceral aneurysms, and the most common are intra-abdominal vessel aneurysms with a sex ratio of 1:4 (male by female) [1,2]. The incidence of splenic artery aneurysm rupture is 1%-18%; the mortality rate is 25%, but it increases to 20%-50% during pregnancy [3,4]. The incidence

of splenic artery aneurysm rupture in the latter half of pregnancy accounts for 65% of all rupture cases because of the increase in female hormone levels or stress on the blood vessel wall [5]. Maternal mortality rate owing to rupture is 75%, and fetal mortality rate is 95% throughout the pregnancy [6]. However, studies on surgical intervention prior to rupture are rare, and many reported on surgical intervention after rupture [7]. Within our scope, there are extremely few studies on the treatment of unruptured splenic aneurysms during pregnancy, and

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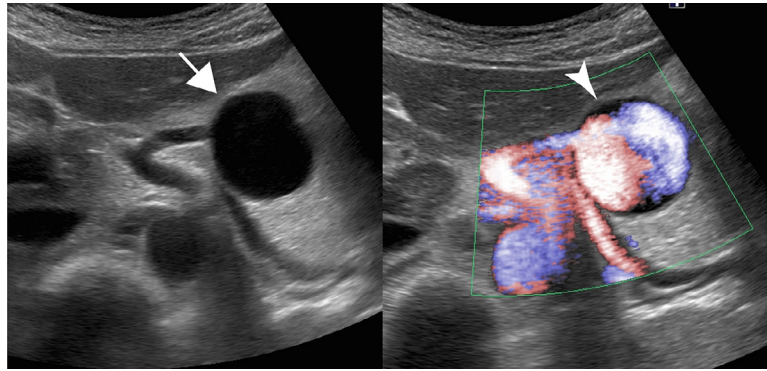


Fig. 1 – Abdominal ultrasonography demonstrates an aneurysm (arrow) with blood inflow signal (arrowhead) with the connection to the splenic artery.

only 1 case was treated with transcatheter arterial embolization (TAE), following which the patient developed splenic abscess [8].

Here, we report a case of a patient who underwent radiation-reducing TAE for proximal side splenic artery aneurysm without any complication at 26 weeks of pregnancy.

Case report

A 30-year-old woman with a history of 1 gestation and 1 pregnancy and without notable medical or family history underwent abdominal ultrasonography (AUS) in a medical checkup, which showed a non-echoic round lesion in the upper abdomen suggestive of pancreatic cyst. After 2 months, she was referred to the department of internal medicine at our hospital. However, during the waiting period, she became pregnant, which was found to be 6-week pregnant. Splenic artery aneurysm was suspected by AUS, and she was referred to the department of gastroenterology on the same day. AUS with color Doppler examination confirmed a 2.8-cm fusiform aneurysm on the proximal side of the splenic artery (Fig. 1). The strategy to treat the patient was discussed in the departments of gastroenterology, obstetrics and gynecology, and radiology. Considering the dangers of radiation exposure to the fetus, TAE was planned for the 26th week of pregnancy, which is thought to be permissible for the fetus given the maturity of the fetus. Additionally, the treatment method was planned as a method involving isolation of the parent artery without intra-aneurysmal embolization to shorten the operative time. Splenic artery aneurysm was evaluated by AUS monthly at the department of gastroenterology; its diameter did not increase until the scheduled treatment day.

To reduce radiation exposure of the fetus, we used a low dose pulsed fluoroscopy, reduced the number of digital subtraction angiographies performed, narrowed the imaging range, and used digital zoom to allow the radiologist to see larger fluoroscopic images without increasing the radiation exposure. A 4.5-Fr shepherd hook-type guiding sheath (Parent Plus 45; Medikit, Tokyo, Japan), was inserted from the bilateral common femoral aorta and advanced into the celiac artery.



Fig. 2 – The angiography from the 4.5-Fr guiding sheath in the celiac artery demonstrates the blood inflow to the splenic aneurysm (arrow).

Angiography revealed the fusiform shape of the splenic artery aneurysm at the proximal side of the splenic artery (Figs. 2 and 3). A 3.8-Fr. High-flow balloon catheter (Pinnacle Blue; Tokai Medical Products, Aichi, Japan) was advanced to the proximal side of the splenic artery aneurysm through a 4.5-Fr guiding sheath. The steerable microcatheter (Leonis Mova 2.4 Fr/ 2.9 Fr; Sumitomo Bakelite, Tokyo, Japan) was advanced to the distal side of the splenic artery aneurysm through a 5-Fr catheter (Imager II; Boston Scientific, Marlborough, MA, USA) from the other side. After advancement of the steerable microcatheter through the 5-Fr catheter to the distal side of the splenic artery aneurysm, the high-flow balloon at the proximal side of the splenic artery aneurysm was inflated to control the blood flow. Then, the distal side of the aneurysm was embolized with Target XL (2 of 8 mm × 30 cm) and Interlock-18 (2 of 6 mm × 20 cm) from the steerable catheter (Fig. 4). After finishing the coil embolization of the distal side of splenic artery aneurysm, the high-flow balloon was deflated and advanced inside the aneurysm, the steerable catheter was pulled out, and the tip of the 5-Fr catheter was placed at the proximal side of the splenic artery aneurysm. After inflating the balloon catheter

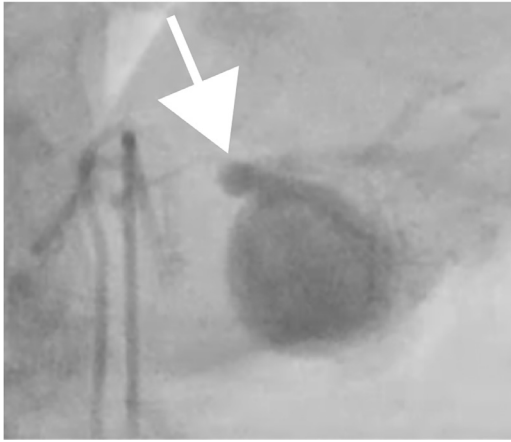


Fig. 3 – The angiography from the 4.5-Fr guiding sheath in the celiac artery taken 0.3 s after [Figure 2](#) demonstrates the blood inflow from the splenic aneurysm to the distal side (arrow).

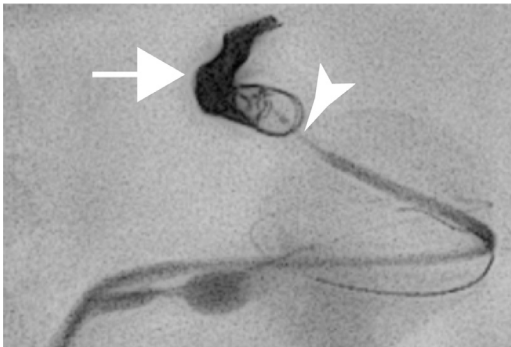


Fig. 4 – The distal side of the splenic artery aneurysm was embolized with coils (arrow) from the steering catheter (arrowhead).

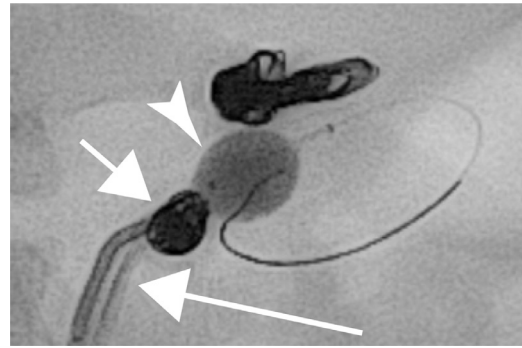


Fig. 5 – The proximal side of the splenic artery aneurysm was embolized with coils (arrow) from the 5-Fr catheter (large arrow) by applying force against the inflated balloon in the aneurysm (arrowhead).



Fig. 6 – Angiography of the celiac artery after finishing isolation to the splenic artery aneurysm demonstrates the preserved blood flow of the distal side of the aneurysm with collateral pathways (arrow).

in the splenic artery aneurysm, inflow of the splenic artery aneurysm was blocked by pulling the balloon catheter and applying force against the orifice of the aneurysm. The proximal side of the aneurysm was embolized with Interlock-35 (6 mm × 10 cm) from the 5-Fr catheter. Additionally, the 2.2-Fr microcatheter (Progreat β^3 ; Terumo, Tokyo, Japan) was inserted in the 5-Fr catheter; then, the Interlock-18 (6 mm × 10 cm) and Galaxy (7 mm × 21 cm, 8 mm × 24 cm) were placed at the proximal side of the splenic artery aneurysm ([Fig. 5](#)). After finishing embolization of the proximal side of the splenic artery aneurysm, angiography of the celiac artery flow to the splenic artery aneurysm showed that the blood flow in the spleen was maintained ([Fig. 6](#)). AUS showed no blood flow signal in the splenic artery aneurysm at that time. The total irradiation dose was 268 mGy. The patient had no complications from the procedure and did not suffer from splenic artery aneurysm rupture until childbirth.

On the 37th week of pregnancy, the patient underwent vaginal birth without any complications. AUS at 6 months after embolization showed a slight blood flow signal in the splenic artery aneurysm ([Fig. 7](#)), and fat-saturated contrast-

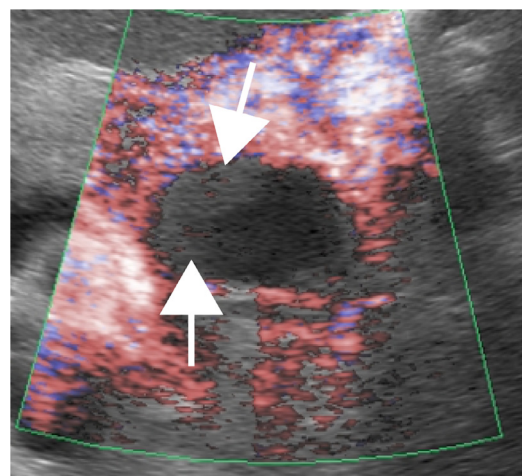


Fig. 7 – Color Doppler of abdominal ultrasonography demonstrates slight inflow signal in the splenic artery aneurysm (arrow).

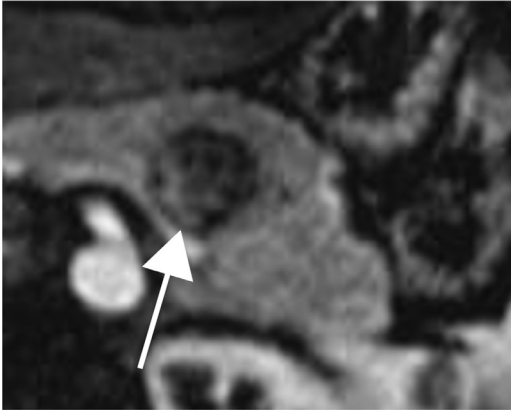


Fig. 8 – The fat-saturated contrast-enhanced gradient echo MRI demonstrates slight enhancement in the splenic artery aneurysm (arrow).

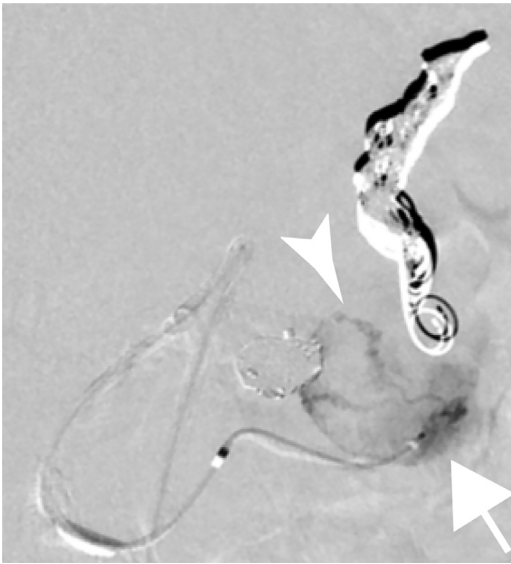


Fig. 9 – The delayed phase of angiography from the microcatheter demonstrates the slight enhancement in the splenic artery aneurysm (arrow) from the collateral pathway (arrowhead) from the transverse pancreatic artery.

enhanced gradient magnetic resonance imaging showed slight enhancement in the splenic artery aneurysm (Fig. 8). The size of the splenic artery aneurysm did not increase compared with that before treatment.

On the second session of TAE at 3 months after delivery, additional embolization was planned. A 4.5-Fr shepherd hook-type guiding sheath (Parent Plus 45; Medikit) was inserted from the right common femoral artery and advanced to the celiac artery. Angiography from the guiding sheath demonstrated a feeding artery to the aneurysm derived from the dorsal pancreatic artery arising from the common hepatic artery (Fig. 9). A triple coaxial system comprised of a 4-Fr catheter (Glidecath; Terumo), 2.8-Fr microcatheter (Renegade HI-FLO; Boston Scientific), and 1.9-Fr microcatheter (Carnelian Marvel

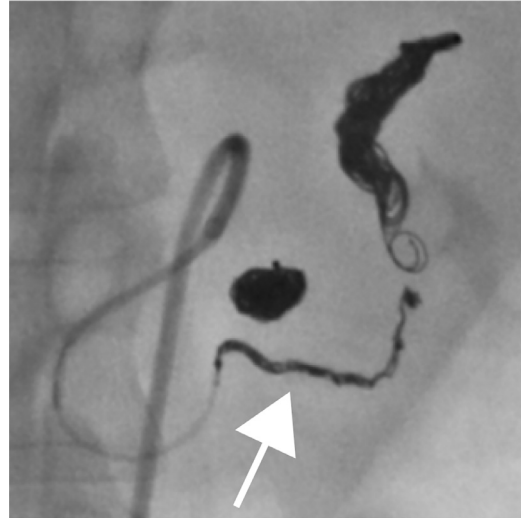


Fig. 10 – Coil embolization of the collateral pathway (arrow) from the transverse pancreatic artery was performed.

NT; Tokai Medical Products) was inserted in the 4.5-Fr guiding sheath and advanced through the feeding artery to the aneurysm. However, it was difficult to access the aneurysm because the feeding artery was small; hence, we planned to perform embolization of the feeding artery by galaxy complex (2.5 × 35 mm, 2.5 × 25 mm) and galaxy helical (2 × 80 mm) (Fig. 10). Final angiography revealed the stoppage of blood flow to the aneurysm and preservation of the distal side of the splenic artery (Fig. 11). No complications were observed on the second session. The blood flow signal of the splenic artery aneurysm on AUS disappeared afterward, and the splenic artery aneurysm became slightly smaller. The patient was followed up 2 years until her moving out; however, the splenic artery aneurysm did not show regrowth contrast enhancement on MRI and collar Doppler signal on AUS.

Discussion

Pregnancy is considered one of the most significant risk factors of the rupture of a splenic artery aneurysm as >400 cases of ruptured splenic artery aneurysms have been reported in the English literature in which approximately 30% of cases developed during pregnancy [2]. The recent guideline for the management of visceral artery aneurysm suggests that splenic artery aneurysm in pregnancy should be treated regardless of its size because of the high mortality rate of both the mother and fetus in case of rupture [9]. Minimally invasive therapy should be employed in the treatment of a pregnant patient with splenic artery aneurysm. Endovascular treatments are less invasive compared to open surgery; however, the problem of fetal radiation exposure exists. It is preferable to perform endovascular treatments after 26 weeks of pregnancy because it is more harmful to the fetus to be exposed to radiation in the early stages of pregnancy (<25 weeks) [10]. Additionally, the risk of rupture of a splenic artery aneurysm

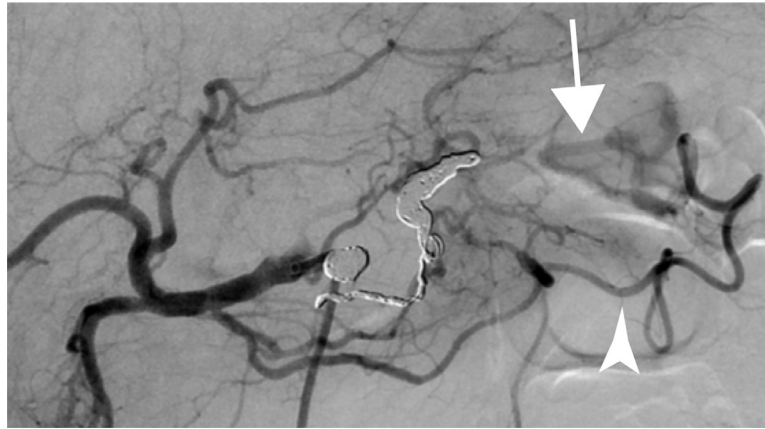


Fig. 11 – The angiography after finishing the coil embolization from the 4.5-Fr guiding sheath in the celiac artery demonstrates no enhancement in the splenic artery aneurysm and preservation of the distal side of the splenic artery (arrow) through the transverse pancreatic artery (arrowhead).

increases after 28 weeks of pregnancy, which accounts for approximately 80% of the ruptured splenic artery aneurysm cases [2]. In this case, considering the risk of rupture of splenic artery aneurysm and the fetal radiation exposure problem, we planned to perform endovascular treatment at 26 weeks of pregnancy by regulating radiation dose during the procedure. The splenic artery aneurysm on AUS was a fusiform aneurysm located proximal to the main trunk of the splenic artery, with 1 inflow artery and 1 outflow artery. The blood flow to the spleen should be preserved in the treatment of splenic artery aneurysm. One treatment option in this case is the use of a stent graft to preserve the blood flow in the parent vessel. However, anticoagulant therapy after the procedure can be a problem due to pregnancy [11]. Moreover, a stent graft for a true visceral aneurysm is not covered by insurance in Japan. The recent guideline states that splenic perfusion should be preserved when embolization of both the proximal and distal sides of an aneurysm located in the proximal or middle third of the splenic artery is performed due to the presence of the collateral artery from the short gastric artery [9]. The diameter of the aneurysm in our case was relatively large, which requires a large number of coils for intra-aneurysmal embolization and prolonged procedure time, resulting in the increase in radiation exposure. Hence, we performed a method involving isolation of the parent artery without intra-aneurysmal embolization to shorten the operative time. As a result, the total entrance skin dose was 286 mGy, and the exposure dose could be sufficiently reduced compared with those in a previous meta-analysis [12]. Additionally, using a balloon catheter together, we succeeded in controlling blood flow, thereby preventing coil migration and embolizing a short area with a small number of coils. While we performed TAE for the feeding artery via the splenic artery aneurysm on the second session after childbirth, we considered it permissible if there is residual blood flow into the splenic artery aneurysm after isolation because it can prevent the rupture of the splenic artery aneurysm by reducing major blood flow into the splenic artery aneurysm to prevent rupture. There is only 1 report, prior to ours, on a patient who underwent TAE for a splenic artery aneurysm at the hilum of the spleen who devel-

oped a splenic abscess after treatment [7]. The TAE of splenic artery aneurysm in the hilum is a risk factor of splenic abscess so that the indication of splenectomy should be considered in this era according to the current guideline (13). To the best of our ability, we were unable to find a report that described a patient with splenic artery aneurysm during pregnancy who was treated with TAE without any complication.

Conclusion

We successfully performed coil embolization of the splenic artery aneurysm with extremely reduced radiation exposure in a patient at 26 weeks of pregnancy. Treatment must be performed to prevent rupture because pregnancy is a risk factor of splenic artery aneurysm rupture that results in high maternal and fetal mortality rates.

Patient consent

Informed consent has been obtained from the patient for publication of the case report and accompanying images.

REFERENCES

- [1] Carr SC, Pearce WH, Vogelzang RL, McCarthy WJ, Nemcek AA Jr, Yao JS. Current management of visceral artery aneurysms. *Surgery* 1996;120(4):627–33 discussion 33–4.
- [2] Abbas MA, Stone WM, Fowl RJ, Gloviczki P, Oldenburg WA, Pairolo PC, et al. Splenic artery aneurysms: two decades experience at Mayo clinic. *Ann Vasc Surg* 2002;16(4):442–9.
- [3] Caillouette JC, Merchant EB. Ruptured splenic artery aneurysm in pregnancy. Twelfth reported case with maternal and fetal survival. *Am J Obstet Gynecol.* 1993;168(6 Pt 1):1810–11 discussion 1–3.

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- [4] Shahabi S, Jani J, Masters L, Cobin L, Greindl J. Spontaneous rupture of a splenic artery aneurysm in pregnancy: report of two cases. *Acta Chir Belg* 2000;100(5):231–3.
- [5] Trastek VF, Pairolero PC, Joyce JW, Hollier LH, Bernatz PE. Splenic artery aneurysms. *Surgery* 1982;91(6):694–9.
- [6] Sadat U, Dar O, Walsh S, Varty K. Splenic artery aneurysms in pregnancy—a systematic review. *Int J Surg* 2008;6(3):261–5.
- [7] Lang W, Strobel D, Beinder E, Raab M. Surgery of a splenic artery aneurysm during pregnancy. *Eur J Obstet Gynecol Reprod Biol* 2002;102(2):215–16.
- [8] Parrish J, Maxwell C, Beecroft JR. Splenic artery aneurysm in pregnancy. *J Obstet Gynaecol Can* 2015;37(9):816–18.
- [9] Chaer RA, Abularrage CJ, Coleman DM, Eslami MH, Kashyap VS, Rockman C, et al. The Society for Vascular Surgery clinical practice guidelines on the management of visceral aneurysms. *J Vasc Surg* 2020;72(1S):3S–39S.
- [10] International Commission on Radiological P Pregnancy and medical radiation. *Ann ICRP* 2000;30(1):1–43 iii–viii.
- [11] Alshawabkeh L, Economy KE, Valente AM. Anticoagulation during pregnancy: evolving strategies with a focus on mechanical valves. *J Am Coll Cardiol* 2016;68(16):1804–13.
- [12] Johnson P, Wong K, Chen Z, Bercu ZL, Newsome J, West DL, et al. Meta-analysis of intraprocedural comparative effectiveness of vascular plugs vs coils in proximal splenic artery embolization and associated patient radiation exposure. *Curr Probl Diagn Radiol* 2020.
- [13] Bjorck M, Koelemay M, Acosta S, Bastos Goncalves F, Kolbel T, Kolkman JJ, et al. Editor's Choice - Management of the diseases of mesenteric arteries and veins: clinical practice guidelines of the European Society of Vascular Surgery (ESVS). *Eur J Vasc Endovasc Surg* 2017;53(4):460–510.