

# **Case Report**

# Successful endovascular treatment of a spontaneous bleeding accessory spleen: A case report

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### АВЅТКАСТ

Accessory spleens are often encountered in radiologic studies and they are not usually associated with symptoms. They could arise from autotransplantation of splenic tissue after splenic trauma or splenectomy (splenosis) [1]. In this case we describe a woman treated for splenectomy 20 years before and subsequently for adhesions, that suffered sudden left upper abdominal quadrant pain, weakness, and pale color. Contrast-enhanced computed tomography revealed free spilling in the abdomen and venous bleeding of a big accessory spleen; thus the patient underwent transcatheter arterial embolization with coils. Due to the 2 previous surgical operations in the splenic loggia, endovascular treatment compared to "open surgery" was the best choice in this case because of determined less complications, a shorter period of hospitalization, and a reduction of health cost.

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# **Case presentation**

A 42-years-old woman was admitted to emergency department of our hospital affected by left upper abdominal pain, weakness, and sweatness. She did not have any symptoms before and denied histories of recent trauma. She had undergone splenectomy for a traumatic rupture 20 years before due to a car accident and 3 years later a second operation for abdominal subocclusions caused by adhesions. After these 2

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Fig. 1 – CECT Venous phase axial (A) and coronal (B) shows an accessory spleen (blue star) with a tenuous active bleeding inside (red circle) and perisplenic liquid fluid spill (green arrow). CECT, contrast-enhanced computed tomography. (Color version of figure is available online).



Fig. 2 – (A and B) Selective DSA of the splenic artery shows 2 hypertrophic artery branches (blue arrows) arising from the residual portion of the splenic artery (black arrow) and feeding the accessory spleen hilus (red star). (C) Selective DSA of residual splenic artery after embolization demonstrates microcoils (green arrows) at hilus with no further opacification of the accessory spleen. DSA, digital subtraction angiography. (Color version of figure is available online).

surgical operations there was no evidence of accessory spleen. The patient had not been clinically followed-up since the previous surgery performed in another hospital. At admission the patient was awake, blood pressure was 114/74 mm/Hg, heart rate 81 beats/min, body temperature 37.3°C. At clinical examination, abdomen was distended, and painful on the left side. Laboratory examinations revealed only anemia (HB level 11.5 g/dL; RBC:  $3.5 \times 10^3$  U/L). US showed an accessory spleen (anteroposterior diameter 50 mm, transverse diameter 45 mm, longitudinal diameter 55 mm) with iperechoic area inside and surrounded by isoipoechoic fluid spill [2]. In order to perform a better characterization of the lesion, a CECT was performed and revealed a little active spontaneous bleeding inside accessory spleen [3] (Fig. 1). The patient was observed for a week at emergency surgery department. For the persistence of symptoms and the low rate of hemoglobin (11 g/dL) a CECT was performed again; a slight increase of perisplenic liquid fluid spill and a splenic persistent active bleeding were detected. Due to the 2 previous surgical operations in the splenic loggia, the surgeon and the interventional radiologist agreed and decided for endovascular management, so a



Fig. 3 – CECT 2 days after embolization demonstrates spleen devascularization (blue star) and reduction of fluid spill (green arrow). CECT, contrast-enhanced computed tomography. (Color version of figure is available online).



Fig. 4 – (A) US 1 month later shows inhomogeneous ipoechoic accessory spleen (green arrow); (B) no blood sign at color-Doppler. (Color version of figure is available online).

diagnostic angiography was performed. Celiac trunk angiography was performed with a 5 Fr vascular introducer (Cook Medical, Bloomington, Indiana), followed by selective catheterization of residual splenic artery by using a cobra catheter (Merit Medical, South Jordan, Utah) and a microcatheter Progreat 2.7 Fr., (Terumo, Shibuya-ku, Tokyo, Japan). Celiac angiogram demonstrated normal branching pattern [4]. Selective digital subtraction angiography of the splenic artery showed 2 hypertrophic artery branches [5] arising from the residual portion of the splenic artery and feeding the accessory spleen hilus [6] (Figs. 2a and b). The microcatheter was then advanced, in selective way, in the hilar branches; no active bleeding was detected, but together with surgeon we decided to embolize both vessels using four microcoils  $4 \times 2$  mm "Tornado" (Cool Medical, Bloomington, Indiana), one microcoil  $4\times 3mm$  "Tornado" and one microcoil  $2\times 6$ mm "Concerto" (Medtronic B. V. Heerlen, The Netherlands) [7] (Fig. 2c). After embolization neither arterial supply nor further symptoms (fever, pain, alteration on laboratory values) occurred. CECT at 2-day follow-up demonstrated hypoperfusion and reduction of fluid spill (Fig. 3). Patient was resigned after 1 week with normal Hb level (13 g/dL) and US at 1-month follow-up detected heterogenous ipoechoic accessory spleen with no blood sign at color-Doppler (Fig. 4).

# Discussion

The pathophysiological origin of the accessory spleen and splenosis is different: in particular, the accessory spleen has an embryonic origin due to the lack of fusion of the mesogastric nodules. Splenosis is due to damage of the spleen with consequent self-diffusion of splenic tissue [8]. Post-traumatic splenosis has a frequency of 67% and various localizations (peritoneum, retroperitoneum, liver, etc.) [9]. Splenosis cores, unlike the others, are more fragile due to a thinner capsule without elastic tissue. Differently from accessory spleens, splenic implants are fed from the surrounding tissue [10].

Spontaneous bleeding from accessory spleens and splenosis is extremely rare. According to Guo-Dong Shan review, only 20 cases have been reported in literature, most of them treated with surgery [11]. In our case, an observational approach had initially been proposed. Lack of improvement in patient's clinical condition and previous surgery in the splenic loggia, permitted a minimally invasive endovascular embolization, in agreement with the surgeon. In this case endovascular approach compared to "Open surgery" determined a shorter period of hospitalization and a reduction of health cost; besides endovascular embolization causes also less complications than surgery [12]. In conclusion, despite some drawbacks and feasible complications (such as: bleeding, splenic abscess, and iatrogenic vascular injury), endovascular embolization is a valuable tool in nonoperative management of patients affected by splenosis hemorrhage [13].

## **Compliance with ethical standards**

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

### REFERENCES

- Fleming C, Dickson E. Splenosis: autotransplantation of a splenic tissue. Am J Med 1976;61:414–19.
- [2] Corvino A, Catalano O. Superficial temporal artery pseudoaneurysm: what is the role of ultrasound? J Ultrasound 2016;19(3):197–201.

- [3] Santamaría G, Vilana R. Accessory spleen: ultrasonographic and tomographic characteristics. Rev Esp Enferm Dig 1992;81:263–6.
- [4] Corvino F, Silvestre M. Endovascular occlusion of pulmonary arteriovenous malformations with the ArtVentive Endoluminal Occlusion System<sup>™</sup>. Diagn Interv Radiol 2016;22(5):463–5.
- [5] Paladini A, Carriero A. SIRT administration in a difficult hepatic vascularization case; 2019. Case Report, Radiology Case Reports p. 103.
- [6] Patel SR, Lowe S. "Accessory splenic artery: a rare cause of upper gastrointestinal bleeding". Cardiovasc Interv Radiol 2017;40(7):1115–17.
- [7] Borzelli A, Paladini A. Successful endovascular treatment of a recurrent giant celiac artery aneurysm; 2019. Case Report, Radiology Case Reports p. 112.
- [8] Bock D, King B. Splenosis presenting as a left renal mass indistinguishable from renal cell carcinoma. J Urol 1991;146:152–4.

- [9] Gi-Hong C, Man-Ki J. Hepatic splenosis preoperatively diagnosed as hepatocellular carcinoma in a patient with chronic hepatitis b: a case report. J Korean Med Sci 2008;23(2):336–41.
- [10] Maki T, Omi M. "Spontaneous hemorrhage from splenic tissue 13 years after total splenectomy: report of a case". Surg Case Rep 2015;1:91.
- [11] Guo-Dong S, Wen-Guo C. "A spontaneous hematoma arising within an intrapancreatic accessory spleen. A case report and literature review". Medicine (Baltimore). 2017;96(41):e8092.
- [12] Cangiano G, Corvino F. "Endovascular treatment of simultaneous iliac and superficial femoral arterial pseudoaneurysms after stenting procedure complications". Vasc Endovasc Surg 2019;53(2):160–4.
- [13] Corvino F, Giurazza F. Safety and effectiveness of transcatheter embolization in the treatment of internal mammary artery injuries. Radiol Med 2018;123(5):369–77.