Gastric arterial bleeding secondary to chronic occlusion of the splenic artery

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Abstract. *The authors present exceptional case of life threatening bleeding from fundic arterial collaterals in the patient with atherosclerotic chronic occlusion of the splenic artery. Haemostasis was achieved by endoscopic injection of n-butyl-2-cyanoacrylate, which was documented by angiography. The course was complicated by splenic embolism. Recurrence of bleeding was not observed during a nine-month follow-up period. That was why no splenectomy was performed until now. The case report illustrates that arteries supplying the spleen in case of splenic artery occlusion can be a source of severe bleeding. These collaterals can be interrupted endoscopically with the risk of arterial embolism in the coeliac trunk region.*

Key words: *splenic artery occlusion, gastrointestinal haemorrhage, endoscopic haemostasis, acrylate glue, splenic infarction, arteriosclerosis*

Cyrany J, Kopáčová M, Rejchrt S, Krajina A, Bureš J. Závažné krvácení ze žaludečních arteriálních kolaterál při chronickém uzávěru slezinné tepny. Folia Gastroenterol Hepatol 2004; 2 (2): 92 - 98.

Souhrn. *Autoři prezentují výjimečný případ život ohrožujícího krvácení z arteriálních kolaterál ve fundu žaludku u nemocného s aterosklerotickým chronickým uzávěrem slezinné tepny. Krvácení se podařilo zastavit endoskopickou injekcí n-butyl-2-cyanoakrylátu, což je dokumentováno angiograficky. Průběh byl komplikován embolizací sleziny. Během devítiměsíčního sledování nedošlo k recidivě krvácení, proto dosud nebyla provedena splenektomie. Kazuistika dokládá, že arteriální spojky zajišťující výživu sleziny při chronickém uzávěru slezinné tepny mohou být zdrojem významného krvácení. Tyto kolaterály mohou být přerušeny endoskopicky s rizikem arteriální embolizace v povodí truncus coeliacus.*

Klíčová slova: *uzávěr slezinné tepny, krvácení do trávicího traktu, akrylátové lepidlo, slezinný infarkt, endoskopické stavění krvácení, ateroskleróza*

Gastric varices - abnormal portosystemic collaterals in patients with portal hypertension - are a relatively common source of bleeding. In contrast, haemorrhage from arterial submucosal collaterals is quite exceptional.

Case report

A 59-year-old man was admitted to another hospital with haematemesis and melaena. He had undergone renal transplantation one year before admission, after three years of regular dialysis therapy. The atherosclerotic process was manifested before by repeated brain strokes and was aggravated by risk factor accumulation: cigarette smoking, hyperlipidaemia, arterial hypertension, renal failure and obesity.

On admittance the patient was on beta blockers, 100 mg acetylsalicylic acid, 10 mg prednisone, 500 mg mycofenolate mofetil and 300 mg cyclosporine A per day. Repeated upper gastrointestinal endoscopies were performed after admittance, submucous fundic vessels (initially assumed to be varices) were identified to be a source of bleeding and were treated

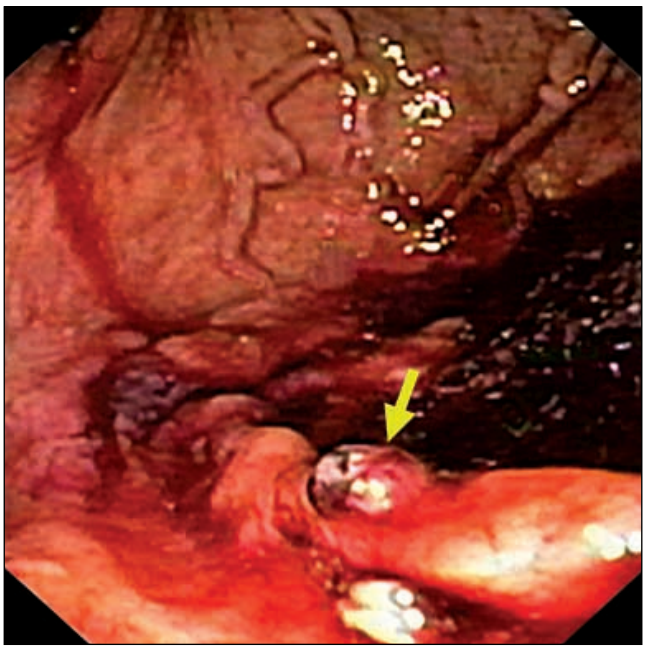

Figure 1
Adherent clot (arrow) is seen on the bulky gastric fold comprising suspected vessel.
Lnoucí koagulum (šipka) je patrné na objemné žaludeční řase tvořené suspektní cévou.

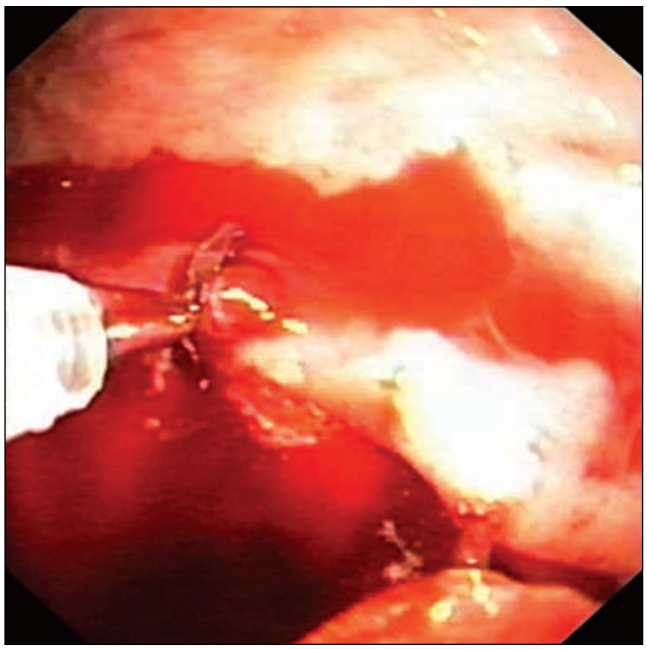

Figure 2
Hemoclip application on the bleeding vessel at gastric fundus
Aplikace hemostatického klipu na krvácející cévu v žaludečním fundu.

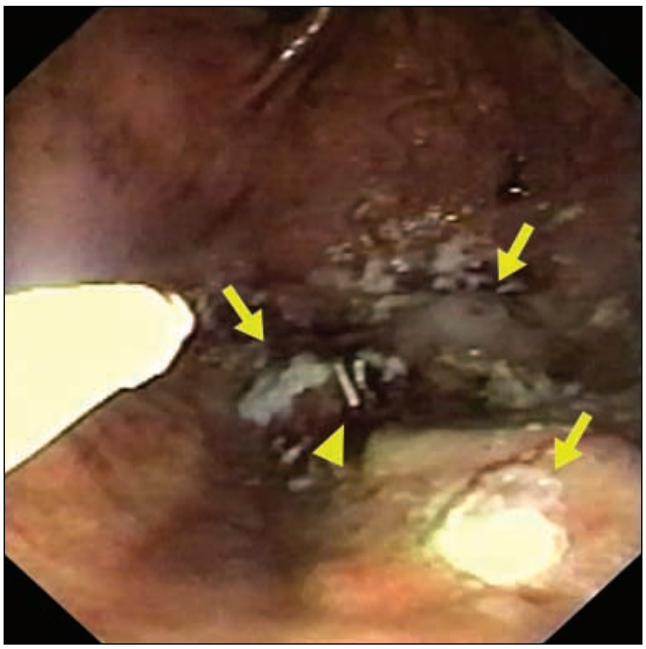

Figure 3
Cyanoacrylate was injected at three different places into the bleeding vessel (arrows). Two hemoclips used for initial control of bleeding (arrowhead) are seen centrally.
Cyanoakrylát byl endoskopicky aplikován na třech místech do krvácející cévy (šipky). Hrot uprostřed obrázku označuje hemoklipy použité k prvotní kontrole krvácení.

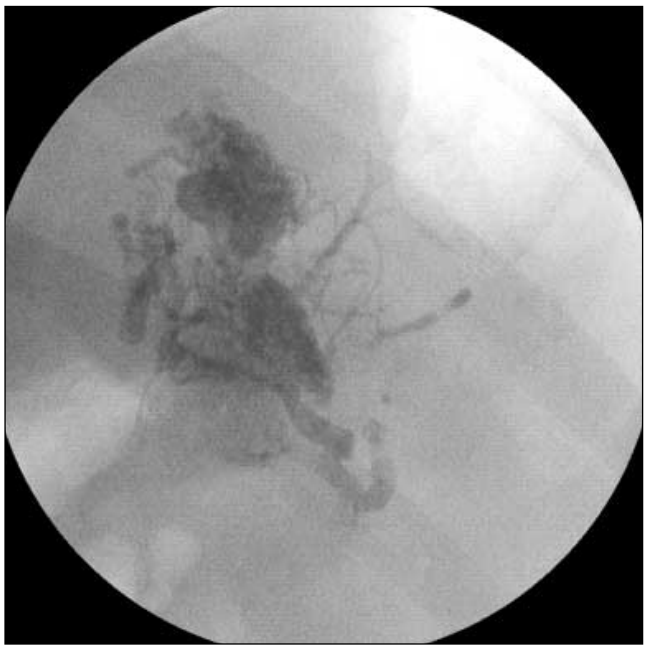

Figure 4
Fluoroscopy image after cyanoacrylate endoscopic injection. Combined submucosal and intravascular application is seen.
Skiaskopický obraz po endoskopické aplikaci cyanoakrylátu. Je patrná kombinovaná submukózní a intravaskulární aplikace.

by injection of 1% polidocanol in an endoscopy unit elsewhere. Terlipressin was administered and mycofenolate was discontinued.

The patient was referred to our hospital for clinical and laboratory signs of on-going bleeding. On admittance he revealed signs of sympathetic activation - he was pale, sweaty, but with normal arterial blood pressure and heart rate, with no clinical signs of chronic liver disease such as hepatomegaly, spider naevi or palmar erythema. Laboratory examination showed normocytic anaemia with values of haemoglobin 82 g/L, haematocrit 0.242, prothrombin time

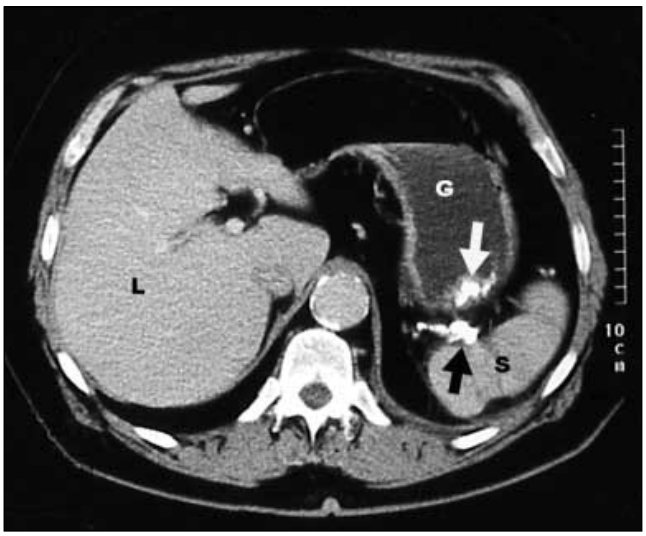

Figure 5
Contrast CT scan two months after endoscopic injection shows acrylate with Lipiodol in the stomach wall (white arrow) and extramurally in the splenic hilum (black arrow). Irregular contour of the spleen (S) reflects previous splenic infarction. L - liver, G - stomach partially filled with water.
Na kontrastním CT snímku dva měsíce po endoskopické aplikaci je patrný akrylát s Lipiodolem ve stěně žaludku (bílá šipka) a extramurálně v hilu sleziny (černá šipka). Nerovný okraj sleziny (S) souvisí s předchozí embolizací sleziny. L - játra, G - žaludek částečně vyplněný vodou.

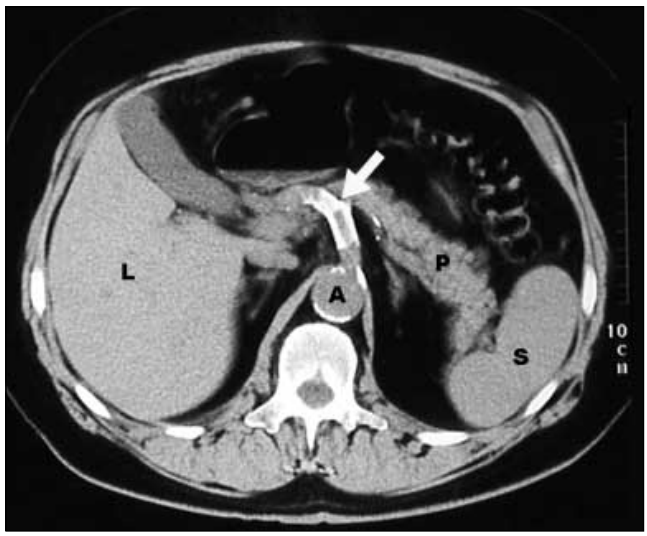

Figure 6
Plain CT scan shows calcifications in the wall of coeliac trunk (arrow) and aorta (A). P - pancreas, S - spleen, L - liver.
Na nativním CT snímku patrny kalcifikace ve stěně truncus coeliacus (šipka) a aorty (A). P - pankreas, S - slezina, L - játra.

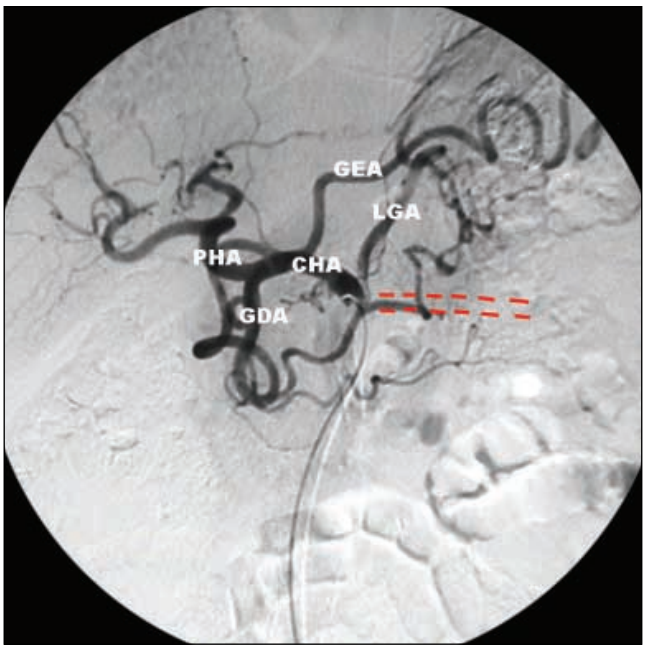

Figure 7
Truncus coeliacus arteriogram. A contrast agent applicated through catheter highlights the common hepatic artery (CHA), the proper hepatic artery (PHA), left gastric artery (LGA) and gastroduodenal artery (GDA) with thickened gastroepiploic arcade (GEA). Obliterated lienal artery marked with red dashed lines.
Arteriogram truncus coeliacus. Aplikací kontrastní látky katetrem je znázorněna a. hepatica communis (CHA), a. hepatica propria (PHA), a. gastrica sinistra (LGA) a a. gastroduodenalis (GDA) se zesílenou gastroepiploickou arkádou (GEA). Obliterovaná slezinná tepna je označena přerušovanými červenými čarami.

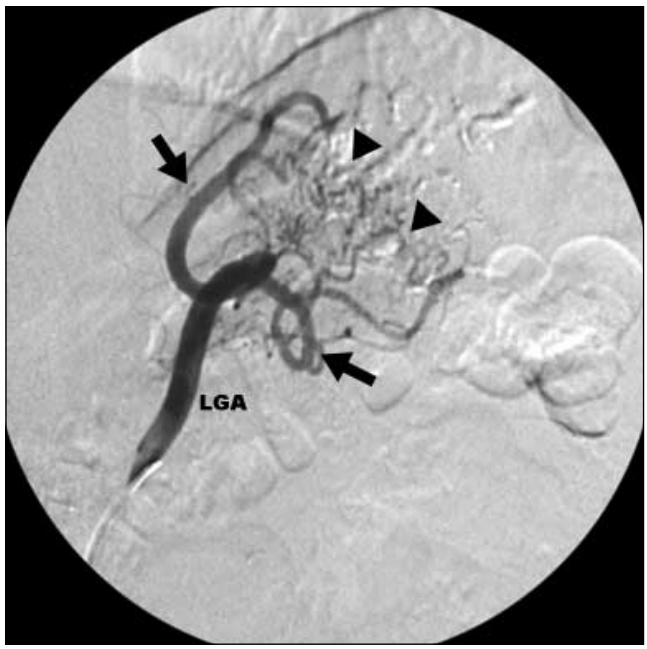

Figure 8
The left gastric arteriogram (LGA) shows thickened arterial collaterals (arrows) partially closed with acrylate (arrowheads).
Na arteriogramu a. gastrica sinistra (LGA) patrné zesílené arteriální kolaterály (šipky) částečně uzavřené akrylátem (hroty).

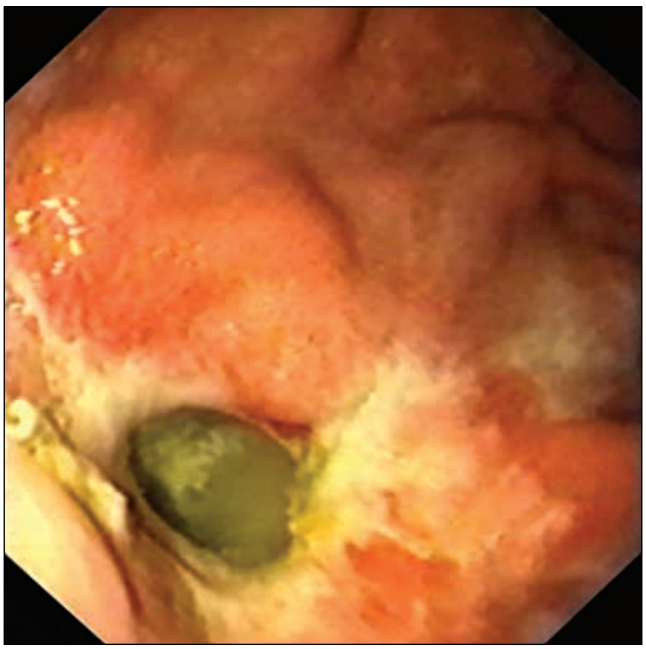

Figure 9
There was a deep ulcer at the upper third on major curvature of the gastric corpus. The ulcer was probably caused by paravascular leak of cyanoacrylate during the application in terrain of previous polidocanol injection.
Hluboký vřed na velké kurvatuře v horní třetině žaludečního těla. Vřed vznikl nejspíše v důsledku paravaskulární aplikace cyanoakrylátu v místě předchozí injekce polidokanolu.

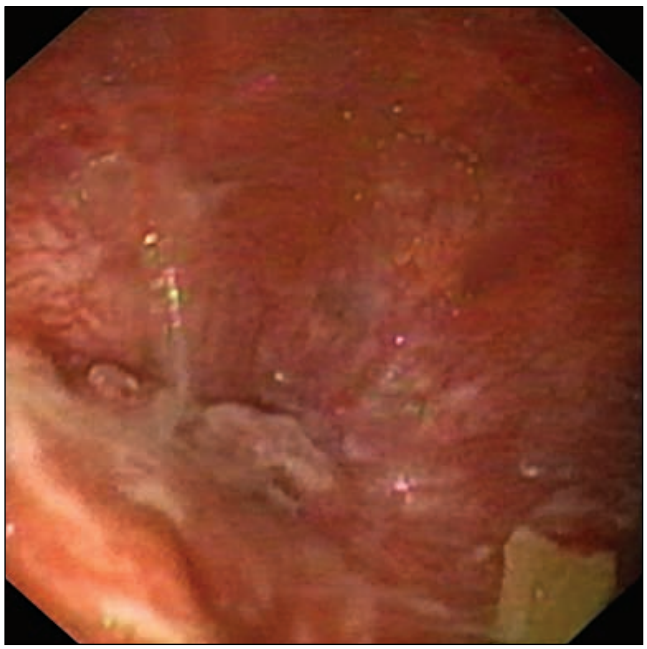

Figure 10
The ulcer seen in Fig. 9 was almost healed within a month.
Vřed (na obr. 9) byl po jednom měsíci téměř zhojen.

1.25 INR, slight creatinine elevation and hydromineral imbalance. Values of bilirubin, aminotransferases, alkaline phosphatase, gamaglutamyltransferase were within normal range.

No signs of actual bleeding were revealed on endoscopy just after transfer of the patient. Marked bulky mucosal folds in the fundus were mentioned, there was ulceration on one of them and signs of previous polidocanol injection were seen.

Two days later, repeated gastroscopy was performed for the recurrence of haematemesis and melaena. After coagulum removal, the source of bleeding was localized to the previously identified small ulceration with adherent clot (Fig. 1) and was classified IIb in the Forrest scale.

The endoscopist decided to treat the lesion with fibrin glue (Tissucol Kit Baxter), but after needle withdrawal spurting pulsatile bleeding appeared and had to be stopped using haemostatic clips (Olympus) (Fig. 2).

Mucosal folds were assumed to be shaped by vascular structures and he decided to inject acrylate glue. Under fluoroscopic monitoring, six doses of acrylate glue (Histoacryl Braun 0.5 mL) mixed with oily contrast agent (Lipiodol Ultra - Fluide, Guerbet, in 1:1 ratio) were applied into vessels underlying the mucosal fold, which the ulcer was located on (Figs. 3 and 4).

The bleeding episode required 11 units of deleukotised resuspended erythrocyte mass and 7 units of fresh frozen plasma until this procedure, after which no signs of ongoing bleeding appeared.

Fourteen hours after the acrylate glue injection, the patient suffered from sharp epigastric pain, ultrasonography in contrast with previous ultrasound examination, demonstrated inhomogenous splenomegaly with focal embolization lesions in the splenic upper pole. Analgesics and antibiotics were administered, subjective complaints, leukocytosis and ultrasonographic finding regressed.

No signs of portal hypertension were found on Doppler ultrasound, invasive measurement of free and wedged hepatic venous pressures also gave normal values - portosystemic gradient was 3 mm of mercury. Free patent trunk and intrahepatic branches of the portal vein were visualized during retrograde carbon dioxide portography, only small reflux to the splenic vein did not allow assessment of its patency. Direct portography was not performed with respect to previous spleen infarction. Computerized tomography showed embolization material in the wall of gastric fundus and also extramurally in the splenic hilum

(Fig. 5). Truncus coeliacus was intensely calcificated (Fig. 6). Splenic, mesenteric and portal veins were patent in the venous phase, unimpaired blood flow in the coeliac trunk and mesenteric artery was seen in the arterial phase. The splenic artery could not be seen.

Coeliac trunk angiography subsequently confirmed previous suspicions. Not even the splenic artery stump was filled with contrast agent (Fig. 7). Chronic splenic artery occlusion was bypassed by huge collaterals from the gastroduodenal artery (via gastroepiploic and inferior pancreatic artery) and from the left gastric artery (via short gastric arteries). Interruption of the last ones by means of previous endoscopic glue injection was seen (Fig. 8), the radiologist did not indicate further endovascular treatment. No signs of arteriovenous fistula were found.

Helicobacter pylori infection was not proved by rapid urease reaction and we found no laboratory signs of hypercoagulation state. Follow-up upper endoscopy found deep mucosal defect (Fig. 9), which was probably caused by paravascular leak of bucrylate during the application in terrain of previous polidocanol injection. The ulcer healed within a month (Fig. 10). In the nine-month follow-up period, no signs of recurrence bleeding were revealed. That is why splenectomy was not indicated so far.

Discussion

Submucosal arterial collaterals, feeding the spleen instead of the closed splenic artery, only caused bleeding in a few cases described so far in available literature. Spriggs (15) reported the case of a 31-year-old man with endoscopic signs of bleeding erosive gastropathy, on whom splenectomy and partial gastrectomy in two steps had to be performed for intractable bleeding. Author guessed the cause to be congenital absence of the splenic artery, like Durans (5) in a similar case report. In the paper published by Baron (1), a 36-year-old patient bled from a peptic lesion localized subcardially on the small curvature. Neither endoscopic bipolar coagulation nor rubber band ligation achieved haemostasis. Embolization was indicated, followed by splenectomy with partial devascularization of the great curvature. The author assumed that the cause of splenic artery occlusion was blunt abdominal trauma 2 years before the first bleeding episode.

Aetiology of splenic arterial occlusion is well documented by Worthley (18). Elective ligation of distal splenic artery with splenic conservation was performed on a 68-year-old woman for an accidentally found aneurysm 13 years before bleeding. Gastroscopy found an adherent clot in the fundus. Angiography and laparotomy found rich collaterals between the proximal splenic artery and the short gastric arteries. Ligation of splenic vessels, the left gastric artery and the short gastric arteries with subsequent splenectomy had to be performed. Prenominat authors did not refer to any other similar case, neither did we.

Table 1 shows what could cause the absence of usual splenic arteriogram in the usual location. The splenic artery can be a branch of the superior mesenteric artery, the left gastric artery, the middle colic artery or even of the abdominal aorta (10). Branching of hepatolienogastric blood supply is very variable, a "textbook case" tripus Halleri (trifurcation of truncus coeliacus in one point) is found in approximately one quarter of cases.

We can not only come across iatrogenic closure not only after surgical, but also endovascular procedures. Indications for splenic artery embolization are: splenic trauma, aneurysms of the splenic artery and arteriovenous fistulas, preparation prior to splenectomy, segmental portal hypertension, splenic artery steal syndrome following liver transplantation or locoregional chemotherapy of pancreatic carcinoma. Although a lot of these procedures are performed, we could not find any other case of bleeding from arterial collaterals after such a procedure but that mentioned above.

Atherosclerotic aetiology of splenic artery occlusion in our patient is evidenced by coeliac trunk calcification in native CT scans, furthermore cigarette smoking, hyperlipidaemia, arterial hypertension, renal failure and obesity together constitute risk factor accumulation. In addition the atherosclerotic process was proved by CT scan also in intracranial arteries and manifested as recurrent brain strokes.

Once the splenic artery is not patent, splenic blood supply is provided through the right gastroepiploic artery (from the gastroduodenal artery), which in most cases made anastomosis with the left gastroepiploic artery on the greater curvature. Apart from other possibilities (16) (Table 2), there are anastomoses between gastric branches of the left gastric artery and the short gastric arteries, it was in this region that bleeding from submucosal arterial collaterals arose.

Acetylsalicylic acid and cortisonoid use, renal

Table 1
Absence of splenic arteriogram in usual location

alternative location
congenital absence
atherosclerotic obturation
thrombotic obturation
iatrogenic sealing (surgical or endovascular)

Table 2
Collateral blood supply in the case of splenic artery occlusion

a. gastroepiploica (a. gastroduodenalis)
aa. gastricae breves (a. gastrica sin.)
rami pancreatici (a. gastroduodenalis, a. mesenterica sup.)
retroperitoneal collaterals (a. mesenterica sup. seu inf.)
a. lumbalis sin.

insufficiency and arteriosclerosis are ulcerogenic biases, mycofenolate mofetil can induce upper gastrointestinal bleeding as well, peptic lesion above submucosal arterial collaterals was the source of dramatic bleeding.

Since the first endoscopic use of n-butyl-2-cyanoacrylate injection in late eighties (14), this technique has become (except the United States) the method of choice for treatment of bleeding subcardial varices (2,13), in which it is according to randomized controlled trials better than rubber band ligation (9) and sclerotherapy (11) and is highly effective in haemostasis of bleeding peptic ulcers (8) and Dieulafoy lesions (3).

Tissue glue based on coagulation proteins, routinely used for the cessation of peptic ulcer bleeding, was used for the treatment of gastric varices bleeding in the Datta study (4) with good results in primary haemostasis. In our case, bleeding from the application site occurred.

Apart from common complications of cyanoacrylate injection (paravascular injection, needle sticking in the varix, needle blockage, adherence of the glue to the endoscope, rebleeding, pyrexia etc.) (12), embolism to the pulmonary artery branches could happen during intravariceal application (systemic embolism occurs only in the presence of right-to-left shunt). On the other hand, life-threatening embolism to branches of the coeliac trunk was described during injection to a non-variceal bleeding source (7, 8, 17). Therapy in our case was complicated by splenic infarction. "Post-infarction syndrome" (fever, abdominal pain, vomiting, adynamic ileus, leukocytosis), which follows therapeutic embolization (6), did not fully develop.

Summary

This case documents, in correspondence with the cited authors, that genesis of arterial collaterals in the stomach wall after the splenic artery occlusion of various aetiology can evoke haemodynamically significant bleeding. Endoscopic treatment by the means of cyanoacrylate injection seems to be a sufficient alternative to splenectomy and surgical interruption of collaterals. Splenic infarction developed as a side effect of treatment, which points out the risk of arterial embolisms in hepatolienogastric blood supply during the endoscopic application of n-butyl-2-cyanoacrylate to a non-variceal source of bleeding.

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