

## SECONDARY ARTERIO-ENTERIC FISTULA: CASE REPORT AND REVIEW OF THE LITERATURE

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**SUMMARY** – Arterio-enteric fistula is a rare, but potentially deadly cause of gastrointestinal bleeding. The disease occurs in two forms: primary as a result of atherosclerotic aortic aneurysm, aortitis, trauma, radiation, tumor invasion or penetrating ulcer, and secondary as a consequence of surgical aortal reconstruction. The clinical manifestation is mostly gastrointestinal bleeding, rarely back pain, fever and sepsis. Computed tomography with contrast medium is the most suitable diagnostic test, however, the diagnosis frequently requires explorative laparotomy. A case is presented of secondary arterio-enteric fistula, found two years after surgical treatment of chronic pancreatitis with pseudocystojejunostomy, which clinically manifested with gastrointestinal bleeding. Although there was strong suspicion of arterio-enteric fistula, the diagnosis was not verified by routine work-up, but only on explorative laparotomy.

Key words: *Intestinal fistula; Gastrointestinal hemorrhage; Anemia*

### Introduction

Arterio-enteric fistula, a communication between an artery and the gastrointestinal (GI) tract, is a rare, but potentially fatal cause of GI bleeding. Manifestation of the condition varies from obscure GI bleeding and unexplained fever attacks to massive hematochezia and death<sup>1</sup>.

A high degree of clinical alertness and suspicion is critical for making an accurate diagnosis, but standard diagnostic procedures are frequently non-confirmatory, so the diagnosis is often not made until surgery. We present a case of secondary aortoenteric fistula in a 45-year-old man with a recent history of abdominal surgery, whose diagnosis was made at laparotomy.

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### Case Report

A 45-year-old man was referred to our Department from another local hospital, where he had been treated for 10 days at surgical unit because of recurrent GI bleeding of unknown origin. On computed tomography (CT) scan made at local hospital, a suspect hematoma was found in the hilum of the spleen.

The patient had chronic pancreatitis of ethylic genesis, which was complicated with a pseudocyst, so he underwent pseudocystojejunostomy in 2008.

At admission, the patient was afebrile, pale, cardiorespiratory stable; red blood cells  $3.1 \times 10^{12}/L$ , hemoglobin 85 g/L and hematocrit 0.26/L. Both ultrasound and endoscopic ultrasound confirmed a process in the projection of the spleen, which could in differential diagnosis be a hematoma or a pseudoaneurysm (Fig. 1A, B). Technetium-99-m labeled erythrocyte scintigraphy showed no sign of bleeding. On day 6 of hospital stay, multi-slice CT (MSCT) angiography was done to reveal active bleeding from tortuous

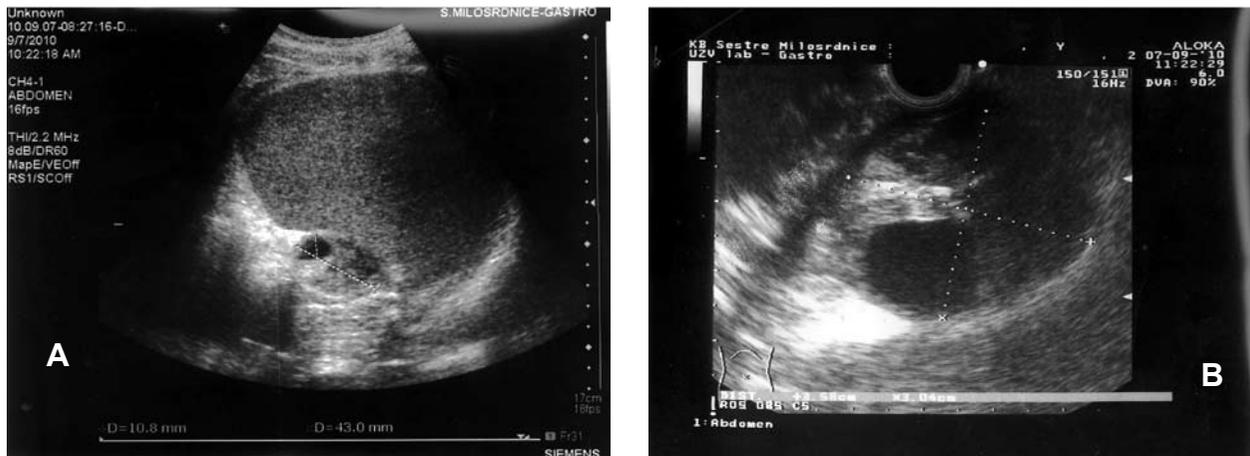


Fig. 1. (A) Abdominal ultrasound; (B) endoscopic ultrasound, both showing a cystic lesion of 4x4.5 cm in size, in the projection of the spleen.

splenic artery into the hematoma formed in the hilum of the spleen (Fig. 2A, B, C). Upper GI endoscopy showed gastric varices. Transarterial embolization of the ruptured splenic artery was attempted, but failed because of the tortuous splenic artery. The treatment was continued at surgical department, where laparotomy was performed. Intraoperative findings were enlarged spleen in contact with hypotrophic left liver lobe and adherent to the stomach, pancreas and posterior abdominal wall. The surgeon disconnected the

cystojejunal anastomosis, resected the splenic aneurysm and detached the spleen from the stomach, pancreas and posterior abdominal wall. Histopathologic examination revealed the spleen with a 5.5-cm section of small intestine attached to it. The slices of small intestine showed bleeding in the bowel wall with small spots of necrosis infiltrated with leukocytes. The operation and postoperative course proceeded without any complications and the patient was discharged from the hospital in good condition.

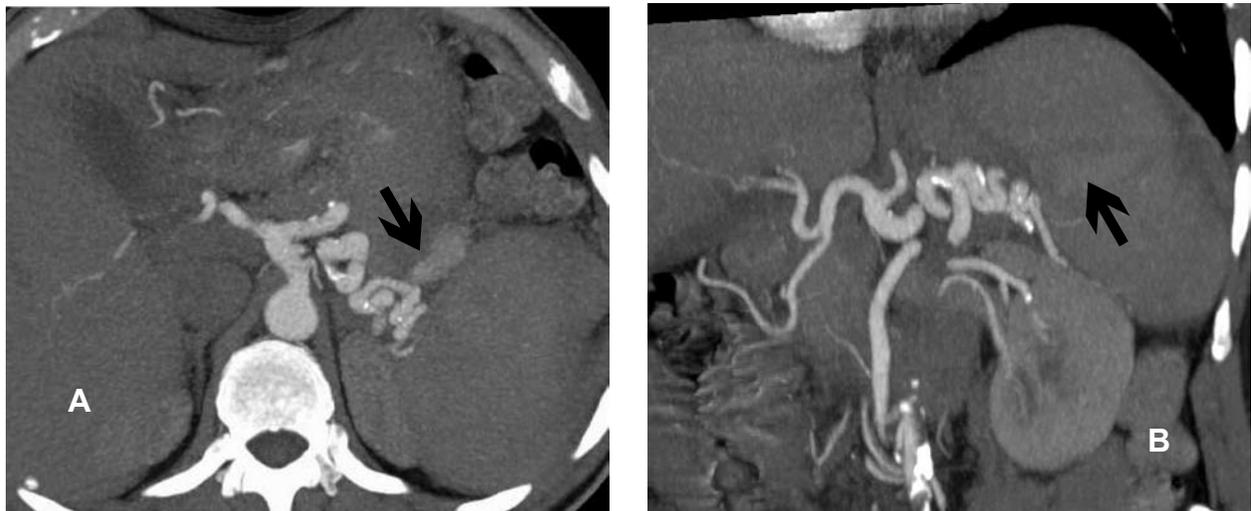


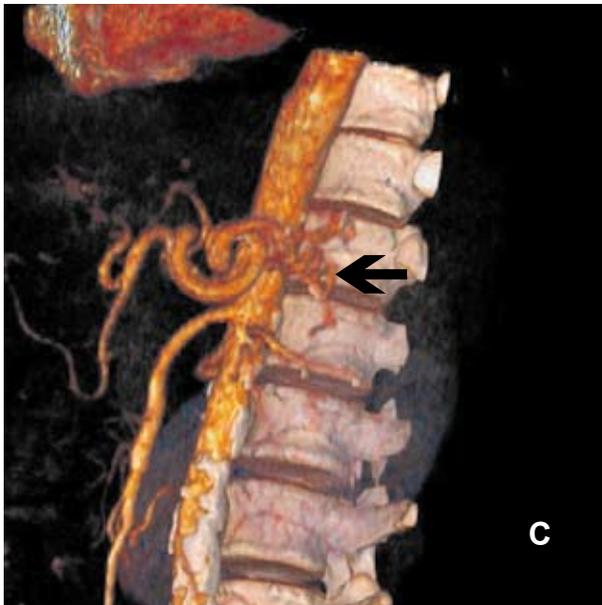
Fig. 2. (A) Computed tomography (CT) angiography axial slice maximum intensity projection (MIP). Arterial phase with active hemorrhage from splenic artery; (B) multislice CT scan in coronary projection; MIP. Active bleeding from splenic artery in the hematoma formed between the tail of the pancreas and the hilum of the spleen; (C) CT angiography with volume rendering (VRT): celiac trunk with tortuous splenic artery.

## Discussion

Aortoenteric fistula (AEF) is a rare cause of acute upper GI bleeding, associated with high mortality if underdiagnosed and not appropriately treated. Aortoenteric, or arterio-enteric fistula is defined as a communication between an artery (most commonly aorta or an artery from the celiac trunk) and GI system. Primary AEF is most commonly a complication of atherosclerotic abdominal aneurysm, prosthesis, radiotherapy, and tuberculosis, whereas secondary AEF usually follows previous arterial reconstructive surgery<sup>2,3</sup>. The incidence of primary AEF ranges from 0.04% to 0.07%, whereas the incidence of secondary AEF has been estimated to 0.4%-2.4%<sup>4-7</sup>.

The most frequent sites of fistula are the third and fourth portions of the duodenum<sup>8</sup>. According to literature reports, primary AEF is often fatal, with the total mortality rate of 80%-100% and perioperative mortality rate of 18%-63%<sup>6,7,9,10</sup>.

The diagnosis and treatment of arterio-enteric fistulas are difficult. The condition manifestation can vary, so a high level of suspicion in predisposed patients is crucial. Often, these patients first present with obscure bleeding, defined as bleeding from the GI tract that persists or recurs without obvious etiology after upper endoscopy, colonoscopy, and radiologic evaluation of the small bowel, which can later be followed by massive bleeding and exsanguinations<sup>11</sup>.



Endoscopy is the procedure of choice for diagnosis and exclusion of other causes of acute upper GI bleeding, but can also sometimes be misleading because a normal finding or a positive finding of gastritis or ulcers without active bleeding does not rule out AEF<sup>12,13</sup>.

Abdominal CT scan and aortography are useful in confirming the diagnosis, and in surgical planning, as well as scintigraphy that can document an active bleeding in the GI tract<sup>14,15</sup>.

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### Sažetak

#### SEKUNDARNA ARTERIJSKO-ENTERIČNA FISTULA: PRIKAZ SLUČAJA I PREGLED LITERATURE

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Arterijsko-enterična fistula je rijedak, ali potencijalno smrtonosan uzrok krvarenja iz probavnog sustava. Bolest se javlja u dva oblika: kao primarna, nastala kao rezultat aterosklerotski promijenjene aneurizme aorte, aortitisa, traume, zračenja, invazije tumora ili penetrirajućeg ulkusa, te kao sekundarna, odnosno posljedica kirurške rekonstrukcije aorte. Klinički se najčešće manifestira u vidu krvarenja iz probavnog sustava, rjeđe bolovima u leđima, vrućicom i sepsom. Najprikladniji dijagnostički test je kompjutorizirana tomografija, no sama dijagnoza se često postavlja tek eksploracijskom laparotomijom. Prikazuje se slučaj sekundarne arterijsko-enterične fistule nađene dvije godine nakon kirurškog liječenja kroničnog pankreatitisa pseudocistojejunostomijom, koja se klinički manifestirala gastrointestinalnim krvarenjem. Iako je postojala velika sumnja na arterijsko-enteričnu fistulu dijagnoza se nije mogla potvrditi standardnim dijagnostičkim postupcima, nego tek na eksploracijskoj laparotomiji.

Ključne riječi: *Intestinalna fistula; Gastrointestinalno krvarenje; Anemija*