

A very rare cause of retroperitoneal bleeding in young patients: do not forget the occurrence of a ruptured left gastroepiploic artery aneurysm!

E. SINAGRA^{1,2}, F. BUSCAGLIA³, A. GIGANTI⁴, V. BONO⁴, D. DI GRUSA³, C. SCIUMÈ^{3,5}

SUMMARY: A very rare cause of retroperitoneal bleeding in young patients: do not forget the occurrence of a ruptured left gastroepiploic artery aneurysm!

E. SINAGRA, F. BUSCAGLIA, A. GIGANTI, V. BONO, D. DI GRUSA, C. SCIUMÈ

Visceral artery aneurysms represent a very rare condition. The affected patients usually present as emergency secondary to the sudden rupture of the aneurysm or as an incidental finding on imaging.

In this setting, gastric and gastroepiploic aneurysms account for only about 4% of all the splanchnic aneurysms.

Since ruptured visceral aneurysms present a high mortality, a prompt and adequate (surgical or radiological interventional) treatment is mandatory.

Due to the difficulty in achieving an adequate transcatheter access in some cases the emergency laparotomy may represent the only chance for the recover of the affected patients.

We report two cases of ruptured left gastroepiploic aneurysms occurred in two young male patients, treated respectively with emergency laparotomy and laparoscopy.

KEY WORDS: Aneurysm - Ruptured - Haemostasis - Left gastroepiploic artery - Emergency treatment.

Introduction

Visceral artery aneurysms represent a very rare condition (1). The affected patients usually present as a clinical emergency secondary to the sudden rupture of the aneurysm or as an incidental finding on imaging (2). Among the splanchnic aneurysms, splenic artery aneurysms are the most common ones accounting for 60% of all splanchnic aneurysms and gastric and gastroepiploic aneurysms account for only about 4% (1, 3-8), thus representing often a

diagnostic dilemma for clinicians, due to their rarity. We report two cases of ruptured left gastroepiploic aneurysms occurred in two young male patients, treated respectively with emergency laparotomy and laparoscopy.

Case report 1

A 30-year-old male patient moved to the emergency unit due to the sudden onset of acute upper abdominal pain and syncope. His medical history was unremarkable. Furthermore, he did not present a history of chest pain, loss of consciousness or breathlessness. His body mass index was 24. He presented with tachycardia (pulse rate of 120/min), and was haemodynamically unstable (supine blood pressure 80/50 mm of Hg). On physical examination there was epigastric tenderness with a tense abdo-

¹ Endoscopy Unit, "Fondazione Istituto G. Giglio", Contrada Pietra Pollastra Pisciotto, Cefalù, Italy

² Euro-Mediterranean Institute of Science and Technology (IEMEST), Palermo, Italy

³ Surgery Unit, Ospedale San Giovanni di Dio, Contrada Consolida, Agrigento, Italy

⁴ School of Medicine, University of Palermo, Palermo, Italy

⁵ DICHIRONS, Dipartimento di Discipline Chirurgiche, Oncologiche e Stomatologiche, University of Palermo, Palermo, Italy

Corresponding author: Emanuele Sinagra;
e-mail emanuelesinagra83@googlemail.com

men. No mass or organ was palpable. His haemoglobin (Hb) on admission was 8 g/dl, and white cell count was 12.1/dl. All other investigations including ECG, chest X-rays and biochemistry were within normal limits. An urgent CT scan, with and without intravenous contrast, showed in the left abdomen, between the stomach, the pancreas and the spleen, the presence of a coarse hypodense, inhomogeneous formation, with a diameter of 14 cm, with HU values between 50-60, which did not assume contrast enhancement if not in small peripheral areas and then central, referable to supplied hematoma. Peripherally, the lesion shows a hyper-representation of mixed vascular type (arterial and venous), with coexistent free collection in the abdomen, in the absence of densitometric changes to the liver, spleen, pancreas, kidneys and adrenal glands (Figure 1). He was given blood transfusions.

The interventional radiologists were consulted, however, due to the hemodynamic instability of the patients, an emergent laparotomy was preferred. At laparotomy, the aneurysm of the left gastropiploic artery was easily identified, though was wrapped in omentum (Figure 2). The artery was ligated on ei-

ther side of the aneurysm and it was excised. He had an uneventful postoperative recovery and was discharged home on the fourth day. One month later, a contrast-enhanced CT scan showed the normal appearance of the splanchnic arterial tree (Figure 3).

Case report 2

A 20-year-old male patient moved to the emergency unit due to the sudden onset of acute upper abdominal pain. His medical history was unremarkable. Furthermore, he did not present a history of chest pain, loss of consciousness or breathlessness. His body mass index was 23. His pulse rate was 90 beats/min, and was haemodynamically stable (supine blood pressure 124/65 mm of Hg). On physical examination there was epigastric tenderness with a tense abdomen. No mass or organ was palpable. His haemoglobin (Hb) on admission was 9.6 g/dl, and white cell count was 9.3/dl. All other investigations including ECG, chest X-rays and biochemistry were within normal limits. An urgent CT scan, with and without intravenous contrast, showed in the gastro-

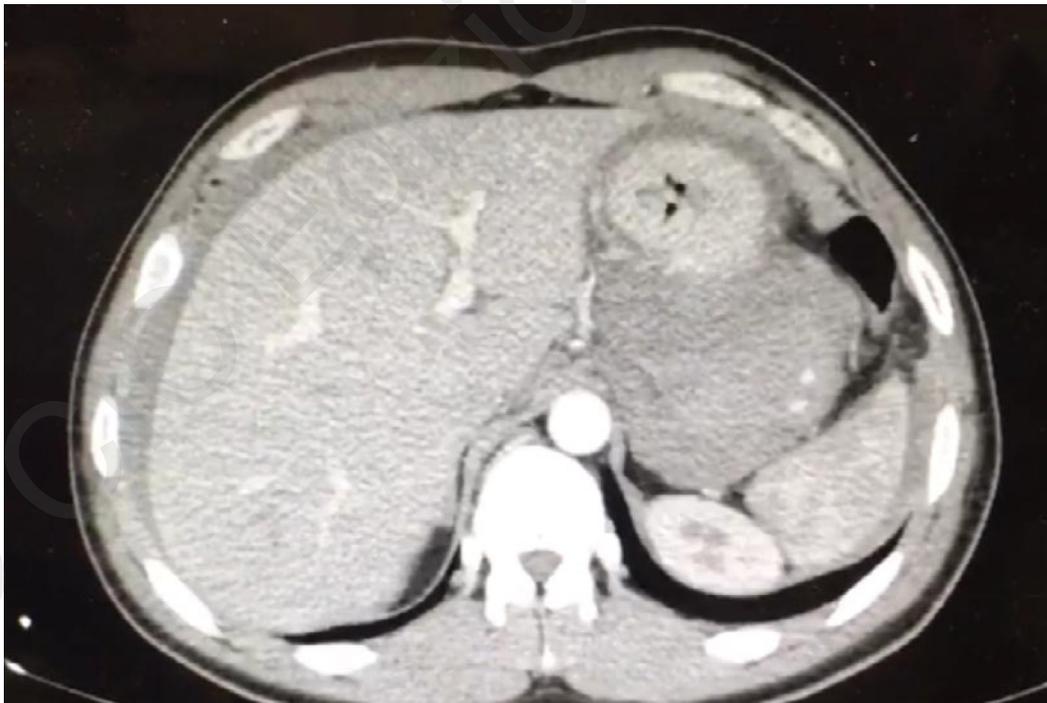


Figure 1 - CT scan, with and without intravenous contrast, showed in the left abdomen, between the stomach, the pancreas and the spleen, the presence of a coarse hypodense, inhomogeneous formation, with a diameter of 14 cm, with HU values between 50-60, which did not assume contrast enhancement if not in small peripheral areas and then central, referable to supplied hematoma.



Figure 2 - The artery was ligated on either side of the aneurysm and it was excised.

splenic space, cranial to the left colic flexure, a tenuously hypodense formation (density 60 HU) of the dimensions of 10 x 7 cm, with polycyclic outlines, showing a contextual blood blush, synchronous with arterial vessels, compatible with blood collection supplied. He was given blood transfusions. The interventional radiologists were consulted, since the patient was hemodynamically stable, and the patient was firstly treated with the embolization of both the splenic and left gastroepiploic artery. However, in the same day of the procedure, a further decrease in haemoglobin and a new occurrence of acute upper abdominal pain were observed. Therefore, an emergency laparoscopy, due to the failure of the interven-

tional radiological treatments, was performed. In this procedure the proximal side of the aneurysm of the gastric artery was clipped using meticulous manipulation. Secondly, the distal side of the aneurysm of the left gastroepiploic artery was carefully exposed from the lesser curvature of the stomach and was clipped, enabling partial resection of the left gastric artery along with the aneurysm to be performed. After the abdominal cavity was irrigated, a drainage tube was placed on the dorsal side of the hepatoduodenal ligament (Figure 4). The macroscopic findings of the resected specimen showed an aneurysm. The patient was discharged from our hospital on day 5 with no postoperative complications.

Discussion

Gastric artery aneurysms represent a very uncommon entity, that account for only about 4% of all the splanchnic aneurysms. They usually occur three times more common in males and in over 50-year-old patients (9, 10).

Usually, they present as solitary lesions occurring due to the result of periarterial inflammation or medial degeneration; in this setting, arteriosclerosis, when present, is thought to be secondary and not a cause (10, 11), and they have also been identified following and described as a complication of acute pancreatitis (12). Since they present with a rupture rate of 90% (8), their commonest clinical presentation is as a surgical emergency with epigastric pain and hypovolaemic shock. Furthermore, when these lesions are intramural (in 70% of cases) they could present with massive haematemesis, while the remaining extramural ruptures (30% of cases) present with intraperitoneal haemorrhage, like in the case reported (13). Since they occur with a mortality of 70% (8), an aggressive fluid resuscitation is necessary while urgent ligation, excision, or embolisation by open, laparoscopic, or interventional techniques is necessary to control the life-threatening bleeding. Few cases, according to the scientific literature, were successfully treated through radiological interventional techniques (1, 10), as well as by using a laparoscopic approach (14). However, a laparoscopic approach in case of ruptured aneurysms could be very difficult due to the massive bleeding that makes not clear the sur-

A very rare cause of retroperitoneal bleeding in young patients: do not forget the occurrence of a ruptured left gastroepiploic artery aneurysm!



Figure 3 - One month after the intervention, a contrast-enhanced CT scan showed the normal appearance of the splenic arterial tree.

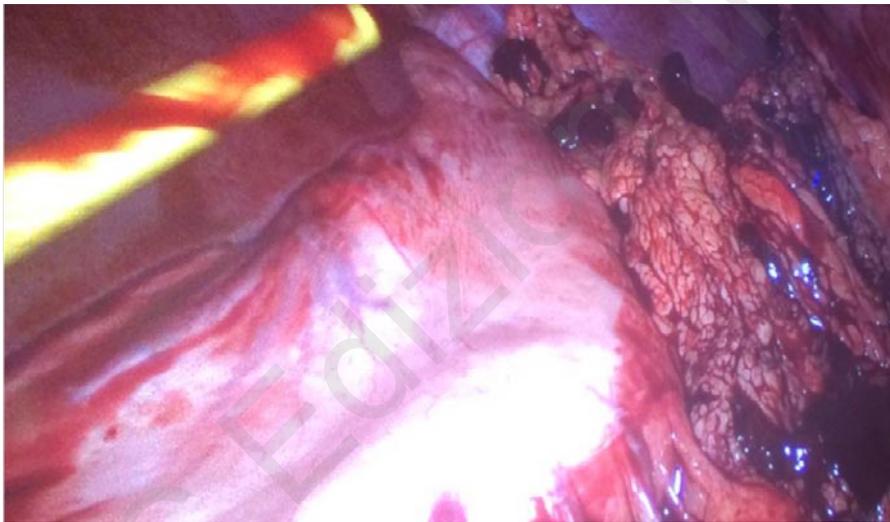


Figure 4 - At the laparoscopy the proximal side of the aneurysm of the gastric artery was clipped using meticulous manipulation. Secondly, the distal side of the aneurysm of the gastric artery was carefully exposed from the lesser curvature of the stomach and was clipped, enabling partial resection of the left gastric artery along with the aneurysm to be performed. After the abdominal cavity was irrigated, a drainage tube was placed on the dorsal side of the hepatoduodenal ligament.

gical field, so it should be avoided as first approach in these cases (15).

We suggest, in hemodynamically unstable patients, an emergency laparotomy (with previous CT scan), with diagnostic and therapeutic purposes; on the other hand, in hemodynamically stable patients, we suggest a CT scan with angiography (and eventual transarterial embolization), followed by a lapa-

roscopic (or laparotomic) approach.

Therefore, to our knowledge, the cases reported represent a very rare cause of retroperitoneal haemorrhage occurring in young male patients, that were successfully treated by emergent laparotomy and laparoscopy, respectively, considering surgery, to date, the mainstay of ruptured left gastroepiploic artery aneurysm.

Conflict of interests

None declared for all the Authors.

Funding support

None.

References

1. Ng CE, Khout H, Farrell R, Fasih T. Left gastric artery aneurysm rupture: a rare cause of retroperitoneal bleeding: a case report. *Scott Med J.* 2016 Aug;61(3):163-166.
2. Carr SC, Mahvi DM, Hoch JR, et al. Visceral artery aneurysm rupture. *J Vasc Surg.* 2001;33:806-811.
3. Rohatgi A, Cherian T. Spontaneous rupture of a left gastroepiploic artery aneurysm. *J Postgrad Med.* 2002;48:288-289.
4. Borioni R, Garofalo M, Innocenti P, et al. Hemoperitoneum due to spontaneous rupture of an aneurysm of the left gastroepiploic artery. *J Cardiovasc Surg (Torino).* 1999;40:63-64.
5. Funahashi S, Yukizane T, Yano K, et al. An aneurysm of the right gastroepiploic artery. *J Cardiovasc Surg (Torino).* 1997;38:385-388.
6. De Angelis M, Vogel C, Horowitz B, et al. Ruptured gastroepiploic aneurysm. *J Clin Gastroenterol.* 1994;18:261-262.
7. Montisci R, Cuccu G, Serra F, et al. Spontaneous rupture of an aneurysm of the left gastroepiploic artery. *Eur J Vasc Surg.* 1992;6:95-97.
8. Stanley JC, Thompson NW, Fry WJ. Splanchnic artery aneurysms. *Arch Surg.* 1970 Dec;101(6):689-697.
9. Stanley JC, Wakefield TW, et al. Clinical importance and management of splanchnic artery aneurysms. *J Vasc Surg.* 1986;5:837-840.
10. Chandran S, Parvaiz A, Karim A, Ghafoor I, Steadman B, Pearce NW, Primrose JN. Ruptured left gastric artery aneurysm successfully treated by thrombin injection: case report and literature review. *The Scientific World Journal.* 2005;5:20-23.
11. James CS, Peter KH. Splanchnic artery aneurysms. In *Surgery: Scientific Principles and Practice.* 3rd ed. Greenfield, L.J., Ed. Lippincott Williams & Wilkins, Philadelphia. 2001, p. 1839.
12. Burke JW, Erickson SJ, Kellum CD, et al. Pseudoaneurysms complicating pancreatitis: detection with CT. *Radiology.* 1986;161:447-450.
13. Witte JT, Hasson JE, et al. Fatal gastric artery dissection and rupture occurring as a paraoesophageal mass: a case report and literature review. *Surgery.* 1990;107:590-594.
14. Toyoda Y, Kyokane T, Kawai T, Watanabe K, Koide F, Ochiai Y, Asai Y, Igami T, Kuze S. Laparoscopic surgery for the treatment of a ruptured aneurysm of the right gastric artery: a case report. *Asian J Endosc Surg.* 2017 Feb;10(1):70-74.
15. Takemoto YK, Fujikuni N, Tanabe K, Amano H, Noriyuki T, Nakahara M. A rare case of spontaneous rupture of an aneurysm of the right gastric artery. *Int J Surg Case Rep.* 2017;40:27-31.