

RUPTURED ILEAL ARTERY PSEUDOANEURYSM: A RARE CAUSE OF LOWER GASTROINTESTINAL HEMORRHAGE

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ABSTRACT

Introduction. Pseudoaneurysms of superior mesenteric artery and its branches are extremely rare but life threatening when complications occur. However, the diagnosis is often missed or delayed because of its non-specific signs and symptoms. Treatments depend on patient's characteristics. In contrast to conventional open surgery, endovascular therapy has been reported as an effective option with minimally invasion, low complication, short recovery time and good outcome, especially in high-risk patients.

Case presentation. We describe a case of ruptured pseudoaneurysm of ileal artery, a branch of superior mesenteric artery, presenting with gastrointestinal bleeding signs while on anticoagulant therapy. Both

Résumé

Rupture du pseudo-anévrisme de l'artère iléale: une cause rare d'hémorragie gastro-intestinale inférieure

Introduction. Les pseudo-anévrismes de l'artère mésentérique supérieure et de ses branches sont extrêmement rares, mais ils menacent la vie lorsque des complications surviennent. Toutefois, le diagnostic est souvent raté ou tardé à cause de ses signes et symptômes non spécifiques. Les traitements dépendent des caractéristiques du patient. Contrairement à la chirurgie ouverte conventionnelle, la thérapie endovasculaire a été rapportée en tant qu'option efficace minimale invasive, à complication réduite, courte récupération et bons résultats surtout chez les patients à haut risque.

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upper and lower gastrointestinal endoscopy were inconclusive. The diagnosis was confirmed by angiography and the patient was successfully treated by coil embolization.

Conclusions. Ruptured pseudoaneurysm of superior mesenteric artery and its branches should be suspected in patients with unexplained gastrointestinal hemorrhage. Endovascular intervention is a feasible approach, especially in patients with multiple comorbidities and high bleeding risk.

Keywords: pseudoaneurysm, superior mesenteric artery, ileal artery, endovascular therapy, coil, gastrointestinal bleeding.

INTRODUCTION

From the histopathological point of view, pseudoaneurysms of visceral arteries are divided into pseudoaneurysms and true aneurysms. Pseudoaneurysm, also known as false aneurysm, is defined as a local enlargement of the artery which is bounded only by the tunica adventitia, the outermost layer of the arterial wall. It is caused by a damage of the artery wall, resulting in the leaking of blood. Conversely, the true aneurysm is an abnormal dilation of an artery involving all three layers of the arterial wall. Therefore, the pseudoaneurysm structure is less stable and more likely to rupture¹. Superior mesenteric artery (SMA) pseudoaneurysm is the third common type, accounting for 5-7% of visceral artery aneurysms¹. It is frequently accompanied by other cardiovascular diseases. Symptoms are variable and not significant. Therefore, most patients are usually diagnosed when the pseudoaneurysm ruptures, leading to high mortality rates. The best strategy to cure is still not known, because of the lack of strong evidence, such as randomized controlled trials. Despite this, endovascular therapy has shown great benefit and is considered the optimal approach².

We report the case of an 84-year-old male patient, under treatment with rivaroxaban, who presented with lower gastrointestinal bleeding from a ruptured pseudoaneurysm of the ileal branch of SMA. The patient was successfully treated with coil embolization. This case reminds a rare cause of gastrointestinal hemorrhage. All the procedures respect the ethical standards in the Helsinki Declaration of 1975, as revised in 2008(5), as well as the national **Présentation du cas.** Nous présentons un cas de rupture d'un pseudo-anévrisme de l'artère iléale, une branche de l'artère mésentérique supérieure, montrant des signes de saignement gastro-intestinale pendant la thérapie anticoagulante. Les endoscopies gastro-intestinales supérieure et inférieure ont été peu concluantes. Le diagnostic a été confirmé par l'angiographie et le patient a été traité avec succès par embolisation par coils. **Conclusions.** Le pseudo-anévrisme rompu de l'artère mésentérique supérieure et de ses branches devrait être soupçonné chez les patients avec de l'hémorragie gastro-intestinale inexpliquée. L'intervention endovas-culaire est une approche faisable, surtout chez les patients aux multiples comorbidités et à risque élevé de saignement.

Mots-clés: pseudo-anévrisme, artère mésentérique supérieure, artère iléale, thérapie endovasculaire, coil, saignement gastro-intestinal.

law. Written informed consent was obtained from the patient.

CASE PRESENTATION

An 84-year-old man presented for three episodes of blood vomiting and five dark red stools. At admission, he had pallor, with altered consciousness. The vital signs were normal. Physical examination was not remarkable, with soft, non-distended and non-tender abdomen, old dysarthria, right hemiplegia because of a prior stroke, and no new neurological signs. His medical history was significant for ischemic stroke caused by atrial fibrillation one month before, treated with transfemoral transcatheter mechanical thrombus aspiration, followed by treatment with rivaroxaban 15 mg per day. Other comorbidities included arterial hypertension, type 2 diabetes mellitus and dyslipidemia.

Laboratory data revealed a hemoglobin of 8 g/dL, blood glucose 325 mg/dL, blood urea nitrogen 26.3 mmol/L, and creatinine 1.07 mg/dL. Coagulation tests, including prothrombin time, partial thromboplastin time, and international normalized ratio were in the normal range. The upper endoscopy did not show any signs of bleeding. The colonoscopy solely detected old blood in the right colon and cecum (Figure 1).

Since both upper and lower gastrointestinal endoscopy were inconclusive, contrast-enhanced computed tomography (CT) was performed and an active bleeding site was observed in the ileum, with SMA as the suspected source (Figure 2). Immediately, angiography was performed, to confirm the diagnosis.



Figure 1. Old blood in the right colon demonstrated by colonoscopy



Figure 3. Digital Subtraction Angiography images. (A) Pseudoaneurysm of the ileal branch of ileocolic artery. (B) Ileal pseudoaneurysm coiling (black arrow).



Figure 2. Abdominal contrast-enhanced computed tomography arterial phase reveals an active bleeding site in the ileum (white arrow).



Angiography identified a pseudoaneurysm of the ileal branch of ileocolic artery, a branch of SMA (Figure 3A). After a careful consideration of all the comorbidities and risks, transcatheter coil embolization was decided. Two OptimaTM coils (1x4 cm and 1.5x4 cm) were inserted successfully to the pseudoaneurysm, to stop the bleeding (Figure 3B). There was no significant blood loss during the procedure. After coiling, the patient recovered and was discharged 7 days after, with no complications.

DISCUSSION

SMA pseudoaneurysm is one of the rarest vascular diseases, with still an insufficient known etiology. It may develop secondary to inflammation (usually from pancreatitis), trauma, infection, vasculitis, or iatrogenic^{1,3}. SMA pseudoaneurysm may remain asymptomatic for years or may rupture. Various factors associated with the risk of rupture have been investigated. In our patient, anticoagulant therapy was possibly the risk factor of rupture. By reducing thrombus formation in the aneurysmal sac, slow and continuous bleeding, anticoagulant treatment could contribute to the enlargement of pseudoaneurysm, which is positively correlated with the risk of rupture. Moreover, anticoagulant treatment also could worsen the severity of bleeding in case of rupture. Since anticoagulants are widely indicated in cardiovascular diseases, further investigation about the impact of these drugs on pseudoaneurysm development and rupture is needed.

Most of unruptured gastrointestinal arteries pseudoaneurysms are asymptomatic or have non-specific symptoms. When rupture occurs, depending on the location of the lesion, bleeding may present as hematemesis, melena, hematochezia, haemobilia or retroperitoneal hemorrhage^{1,4}. Both intra- and extraperitoneal bleeding is lethal, with a mortality rate up to 37%, and requires emergency intervention⁵. However, because of its rarity and atypical clinical manifestations, the diagnosis is challenging, resulting in late treatment and poor outcome. Indeed, in our case, the patient presented with hematemesis and melena, which firstly suggested an upper gastrointestinal bleeding. Hence, other diagnostic techniques are only considered after both upper and lower endoscopy is unable to detect the source of bleeding. CT scan with contrast substance is the most sensitive non-invasive diagnostic method in this case, offering a detailed assessment not only of abdominal organs and their adjacent structures, but also of vascular abnormalities. Therefore, beside angiography, it is considered as an alternative for detecting vascular diseases, such as pseudoaneurysms and aneurysms¹. For this reason, most of true and false aneurysms are detected by CT and confirmed by angiography^{3,6}.

The therapeutic options for pseudoaneurysms include open surgery, laparoscopic surgery, endovascular therapy, or a combination approach⁷. However, the best strategy is still unknown, because of the lack of strong studies comparing the efficacy of these therapies. Therefore, the treatment must be individualized, based on pseudoaneurysm's anatomical characteristics, severity, patient's comorbidities, risk factors, as well as the facility of each center⁸. Recently, endovascular techniques, including embolization, coiling, and covered stents, have been widely preferred, with high rate of success². Various evidence from case reports and series suggests that endovascular therapy is less invasive, safer, and more effective, especially in selected patients with intraluminal hemorrhage, high intra and/or perioperative risks⁹⁻¹¹. It is also highly recommended as the first-line approach for patients with feasible anatomy⁸. Recently, a systematic review and meta-analysis conducted by Barrionuevo et al. indicated that endovascular strategy is more beneficial, with shorter length of hospital stay and lower cardiovascular complications rates¹². Our patient had a high surgical risk, so the endovascular approach was the best option. He recovered without complications.

CONCLUSIONS

Pseudoaneurysms of SMA and its branches should be considered in patients with occult and obscure gastrointestinal hemorrhage. The team approach, including interventional radiologists, gastroenterologists, and radiologists, plays a pivotal role in the management of this pathology. Endovascular treatment is safe, effective and may become the first-line therapy in the future.

Author Contributions:

C.T.C., P.N.K., T.N.M., were responsible for the diagnostic procedures, clinical diagnosis, and treatment decisions. T.N.M performed the endovascular intervention. P.N.K. and D.T.P. wrote and revised the manuscript. All authors have read and agreed to the published version of the manuscript.

Compliance with Ethics Requirements:

"The authors declare no conflict of interest regarding this article"

"The authors declare that all the procedures and experiments of this study respect the ethical standards in the Helsinki Declaration of 1975, as revised in 2008(5), as well as the national law. Informed consent was obtained from the patient included in the study"

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