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Acute acalculous cholecystitis as a rare manifestation of chronic mesenteric ischemia. A case report



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ABSTRACT

INTRODUCTION: Symptomatic chronic mesenteric ischemia (CMI) is an uncommon condition that usually presents with intestinal angina, sitophobia and unintentional weight loss. Acute acalculous cholecystitis (AAC) has very rarely been described in the settings of CMI.

PRESENTATION OF CASE: We describe a case of a 73 year old man that developed an AAC as a complication of CMI. The patient underwent a simultaneous cholecystectomy and open aortic revascularization which was successful. At 24 months of follow-up the patient is clinically well and regained weight.

DISCUSSION: Ischemia has been considered an important etiology for the development of AAC. In the settings of CMI, an AAC might develop has a herald sign of progression to acute mesenteric ischemia and infarction, as the cystic artery is a terminal artery with no collateral network. Performing the aortic revascularization simultaneously with the cholecystectomy might prevent this possible fatal outcome.

CONCLUSION: This case reinforces aortic and visceral occlusive disease as a possible risk factor for the development of AAC, and discusses the treatment controversies when managing both conditions simultaneously.

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1. Introduction

Chronic Mesenteric Ischemia (CMI) is an uncommon condition that is being diagnosed more frequently as the use of imaging tests as CTA became more widespread [1]. Symptoms usually appear when at least two of the three main aortic visceral arteries are occluded [2], and in symptomatic patients treatment is required because of a greater risk of bowel infarction [3]. Symptoms of CMI usually include intestinal angina, sitophobia, and unintentional weight loss [4]. Cholecystitis is very rarely described in the settings of CMI [5,6].

We describe a case of an acute acalculous cholecystitis (AAC) which developed in the context of CMI and discuss its management.

2. Presentation of case

A 73 year old male was referred to our Vascular Surgery Department with a diagnosis of CMI. The patient had been complaining of increasing abdominal epigastric pain over the previous 3 months.

The pain was continuous and persistent but worsened severely after eating and was associated with anorexia and weight loss (20 kgs). He also complained of constipation alternating with diarrhoea and 3 episodes of postprandial vomiting with some relief of the epigastric pain. The patient had a history of hypertension, chronic alcohol consumption (125 g/day), previous smoking habits (stopped 25 years ago), and bilateral lower limb intermittent claudication, accompanied by paraesthesia of the right foot for the last 6 months, when walking. His regular medication included pentoxifylline and telmisartan plus hydrochlorothiazide. On admission, the patient presented with mild epigastric tenderness, weak femoral pulses and absent popliteal, posterior tibial and dorsalis pedis pulses bilaterally.

An abdominal and pelvic CT angiography (CTA) showed diffuse aortic atheromatosis with extensive aortoiliac calcifications, preocclusive celiac trunk stenosis with distal vessels patent, a 2 cm occlusion from the origin of the superior mesenteric artery (SMA), occlusion of the inferior mesenteric artery, left common iliac artery stenosis and occlusion of the right superficial femoral artery (Fig. 1). The patient was scheduled for elective revascularization.

On the 4th day after admission, he referred worsening of his abdominal pain associated with right hypochondrial pain and a positive Murphy's sign, followed by fever, leukocytosis, raised C reactive protein (22,5 mg/dL); aspartate aminotransferase (53 iU/L); alanine transaminase (99 iU/L) and gamma-glutamyl

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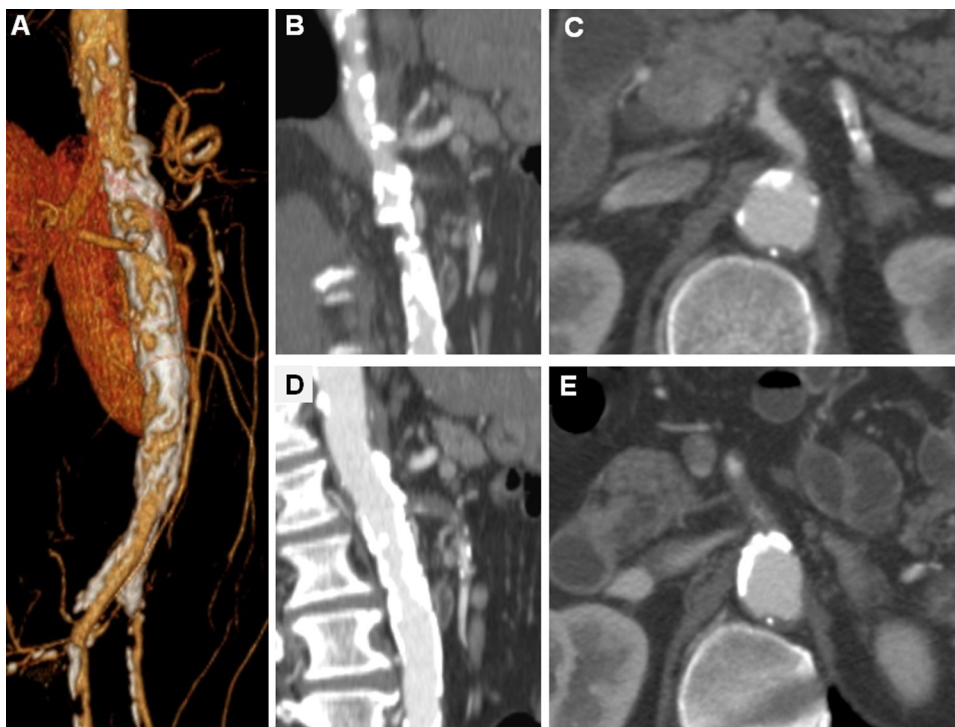


Fig. 1. Pre operative 3D contrast-enhanced CT angiography: lateral view of aorta and origin of visceral vessels, showing preocclusive celiac trunk stenosis and occlusion of the origin and initial 2 cm of the SMA (A). Pre operative contrast-enhanced CT angiography: lateral view of the origin of the celiac trunk (B); cross view of the origin of the celiac trunk (C); lateral view of the origin of the SMA (D); cross view of the origin of the SMA (E).

transpeptidase (295 iU/L). An abdominal ultrasound showed an enlarged gallbladder, with wall thickening and peri-vesicular fluid with no visible calculi, leading to the diagnosis of AAC. The patient was started on piperacillin plus tazobactam.

As there was no improvement over the next 24 h, the patient underwent median laparotomy which confirmed the acute cholecystitis but revealed no loss of integrity of the gallbladder wall or signs of peritonitis. A cholecystectomy was performed, followed by a simultaneous revascularization procedure for treatment of his extensive aortic and visceral occlusive disease. Due to the exuberant calcification of the visceral aorta, precluding infrarenal clamping and anastomosis, a bypass to the common femoral arteries had to be performed from the supraceliac aorta (using a 16 × 8 mm Dacron graft) which was tunneled through the retro-pancreatic area. Visceral revascularization was achieved by retrograde bypasses from the aorto-bifemoral graft to the SMA and to the common hepatic artery (using 8 mm Dacron grafts). (Fig. 2A) After declamping, an area of bowel infarction was recognized in the ileum, requiring a 25 cm small bowel resection and intestinal continuity ensured through mechanical latero-lateral enteric anastomosis. (Fig. 3).

The immediate post-op was uneventful except for a biliary fistula secondary to a cystic stump dehiscence which was resolved by endoscopic sphincterotomy and placement of a prosthesis in the common bile duct. The patient was discharged on the 17th day after surgery.

The post-operative CT angiography showed good patency results with no surgical complications (Fig. 2B). The patient regained weight and is clinically well 24 months after surgery.

3. Discussion

AAC is a rare condition, representing only 10% of all acute cholecystitis and usually appears in critically ill patients, as in the context

of trauma, shock, sepsis and major burns and is associated with a high complication and mortality rate [7]. In the setting of CMI, AAC has seldom been reported in the past as the most common complication of CMI is acute mesenteric ischemia and bowel infarction [1]. Savoca et al. reported two cases where AAC secondary to mesenteric graft occlusion developed in patients who had been treated for CMI [6], and Koea et al. reported a case of AAC in a patient who was later found to have CMI [5]. Although extremely rare, in a seven year study on AAC, Savoca et al. found that 72% of outpatients who developed AAC had clinically significant atherosclerotic vascular disease with no other major conditions to predispose to AAC, which may reinforce aortic atherosclerosis as a risk factor for the development of AAC [8].

Therefore, although AAC has been associated with critically ill patients, recent reports show that AAC can present *de novo* in an outpatient setting without any major illness or associated trauma [8–10], and aortic occlusive disease may represent the most relevant etiological factor in these patients [8].

Currently, AAC is considered to be a multifactorial process, in which bile stasis and ischemia play a major role [7]. Ischemic injury is crucial in the pathogenesis of AAC because the cystic artery is terminal, making the gallbladder susceptible to low-flow states [11]. Furthermore, Hakala et al. reinforced the importance of microcirculatory disturbances in the pathogenesis of AAC [10] and Warren has shown that in contrast to calculus cholecystitis, in AAC, gallbladders show “multiple arterial occlusions with absent or minimal venous filling” [12].

AAC has also been described in a few case reports occurring just before the development of mesenteric infarction in the context of acute mesenteric ischemia [13–15]. In all of these cases, the AAC was thought to be a herald sign of critical ischemia and mesenteric infarction, which resulted in death in two of these case reports [14,15].

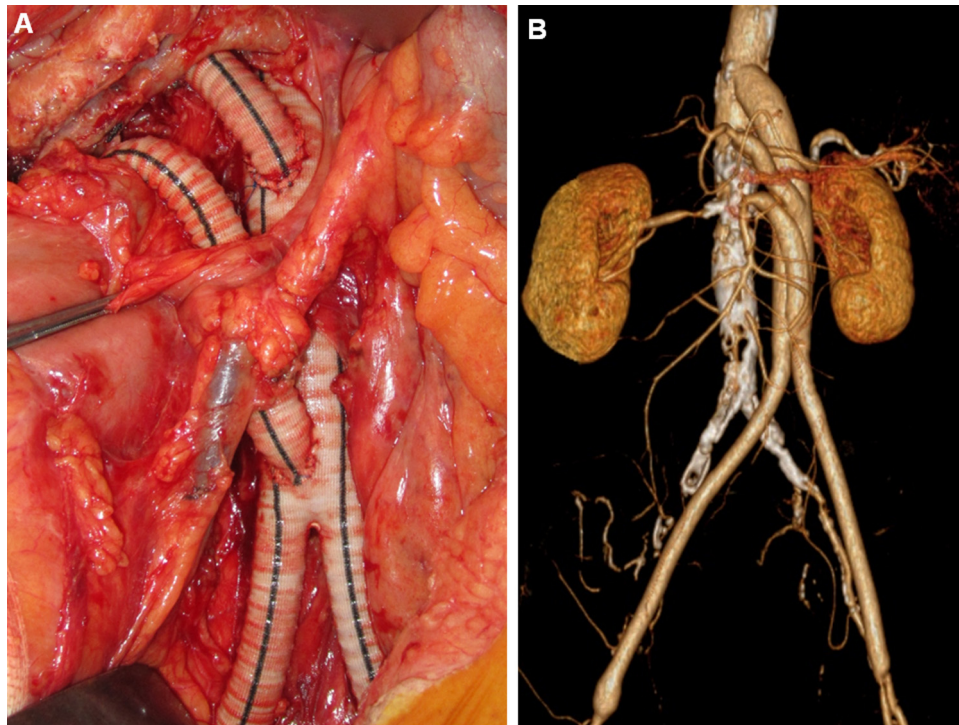


Fig. 2. Aortic revascularization using Dacron grafts showing the supraceliac to bifemoral bypass and retrograde hepatic and superior mesenteric bypasses from the aortobifemoral graft (A). Post operative 3D contrast-enhanced CT angiography showing good patency results (B).

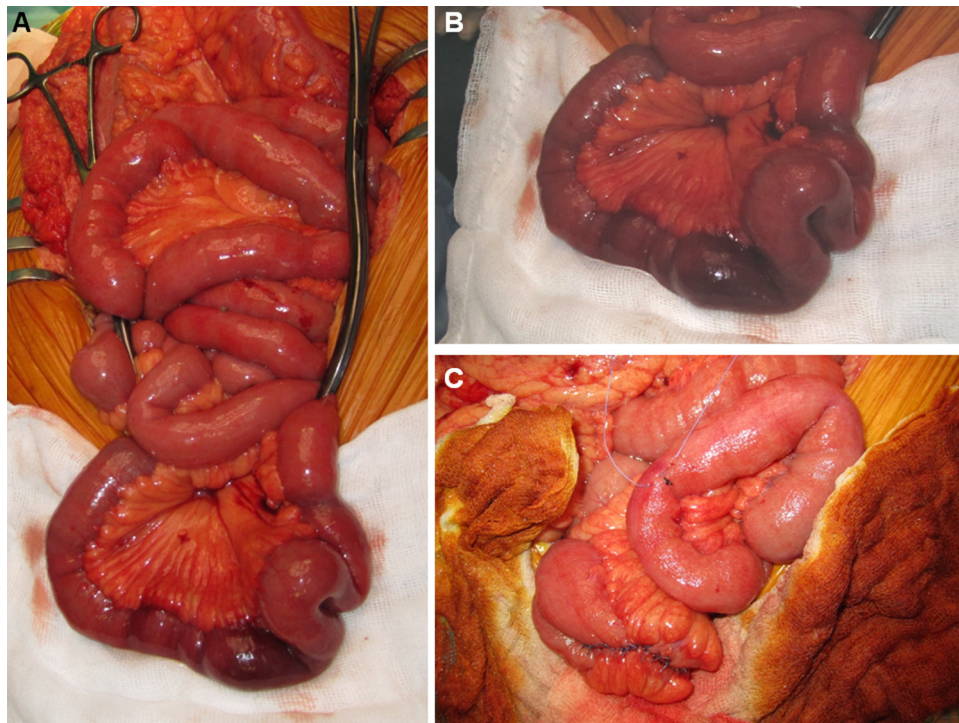


Fig. 3. Area of bowel infarction in the ileum (A) and (B). Post resection mechanical latero-lateral anastomosis (C).

In our case, the AAC might have developed in the same fashion as a consequence of an acute-on-chronic mesenteric ischemia.

AAC management usually includes broad-spectrum antibiotics covering enteric organisms, cholecystectomy and/or cholecys-

tostomy when the patient is critically ill [16]. In this case, because the AAC developed in the context of CMI, a cholecystectomy was indicated [7], raising several questions regarding visceral revascularization, its modality and timing for its performance.

Endovascular mesenteric revascularization has a recognised lower peri-operative morbidity and mortality rate, being particularly appropriate for limited occlusive lesions [1,3]. However, in cases of symptomatic, diffuse aorto-iliac occlusive disease with exuberant calcification of the involved arteries its long-term durability and patency are less favourable when compared with open surgical reconstruction which, in such a setting should be considered the procedure of choice [1,3]. The use of prosthetic grafts for these reconstructions, in a potentially infected territory, following cholecystectomy or bowel resection, has a necessarily higher risk of graft infection. However autologous conduits are rarely available for such extensive aorto-iliac reconstruction. The use of deep veins such as described in the Neoortoiliac System (NAIS) procedure, although feasible, is a very extensive and complex procedure usually not adequate if an urgent revascularization is to be performed in an acutely ill patient [17].

Antibiotic-bonded and antiseptic-impregnated grafts and cryopreserved allografts have been used not only as a preventive technique to avoid future infections but also for *in situ* replacement for infected grafts [18–21]. However, these grafts, are not widely accessible in our institution for urgent settings, and the literature has not shown a clear advantage over non antibiotic/antiseptic coated grafts in potentially infected territories [18,19].

The risk of graft bacterial contamination can be minimized by careful, adequate and meticulous surgical technique [21].

In our case, we used the retro-pancreatic area for tunnelization of the main body of the bifurcated graft, in order to try to reduce the risk of direct contact of the prosthetic material with the bowel and the potentially infected territory. To date and to our knowledge, this technique has not been clearly described in the literature for this purpose and our decision was empirical, in an emergent setting.

Considering the treatment strategy and timing of the procedures, as a cholecystectomy was mandatory, the solution to reduce the risk of graft infection deferred visceral revascularization within a two stage operation could be envisaged. However, if AAC would be considered as a manifestation of visceral ischemia secondary to reduced critical mesenteric perfusion, a higher risk for the development of bowel infarction must be considered, as reported previously [13,14] Therefore, the decision was taken to proceed with immediate mesenteric revascularization in order to minimize the risk and extension of a possible bowel infarct.

The post-operative period was complicated by a biliary fistula treated by endoluminal sphincterotomy without evidence of overt peritonitis or signs of graft infection.

The patient has been well, regained weight, is fully asymptomatic without any clinical or laboratorial evidence suggesting any infectious complication at a follow-up of 24 months.

4. Conclusion

This case reinforces the importance of visceral ischemia as a risk factor of AAC, which can be a possible complication in CMI patients and a marker of severe critical visceral ischemia and a higher risk of progression to acute mesenteric infarction.

Conflict of interest

None.

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We declare we had no financial or other support from industry.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

All of the authors were responsible for the development and revision of the case report.

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Aknowlegements

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The present case report has been reported in line with the CARE guidelines as published in “Gagnier J., Kienle G., Altman D.G., Moher D., Sox H., Riley D.S., and the CARE group. The CARE guidelines: consensus-based clinical case report guideline development. *Journal of Clinical Epidemiology*, 67 (1) 46–51”.

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