RESEARCH ARTICLES

Acute revascularization syndrome after successful treatment of chronic mesenteric ischemia.

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Abstract

Chronic mesenteric ischemia (CMI) is a rare condition, usually caused by stenosis or occlusion of two visceral arteries. As its early symptoms are unspecific, this condition can be diagnosed late or misdiagnosed. Duplex ultrasound or CT angiography or MR angiography is available to reach the diagnosis. Open surgical or endovascular revascularization are two valid options in CMI, even if there is not an evidence-based technique recommended for its intervention. Morbidity and 30-day mortality differ based on the option chosen.

We discuss a case of 59-year-old woman referred for chronic post prandial abdominal pain and weight loss. A computed tomography angiogram (CTA) of the chest, abdomen and pelvis with multiplanar and centerline reconstructions of the mesenteric vessels revealed a multivessel splanchnic mesenteric occlusive disease. We treated her with hybrid revascularization: a retrograde iliac-SMA bypass was performed using a 6-mm diameter PTFE graft end-to-side with SMA and a 5x12mm balloon-expandable stent was inserted into the main trunk of the IMA, via retrograde IMA access.

After the second post-operative day, the patient developed several complications: chest pain associated with dyspnea, abdominal pain with diarrhea and melena, oliguria, sinus tachycardia and laboratory tests's alterations (anemia, neutrophilic leucocytosis, thrombocytopenia, hyperbilirubinemia, increase in transaminases, troponin I and serum creatinine).

A CT scan showed a normal perfusion of the aortic-SMA by-pass and a normal perfusion of the IMA stenting. Blood smear examination revealed presence of schistocytes. The etiopathogenesis was probably a thrombotic microangiopathy due to revascularization of both SMA and IMA.

The patient was successfully treated with high volume fresh plasma infusion and red blood cells units.

For those patients an early diagnosis should be considered, together with an aggressive treatment in the case of occurrence of a systemic inflammatory response syndrome. We therefore suggest to treat these patients only in centers which have the availability of multidisciplinary monitoring and adequate treatment options.

No cases of revascularization syndrome after treatment for CMI are reported in literature.

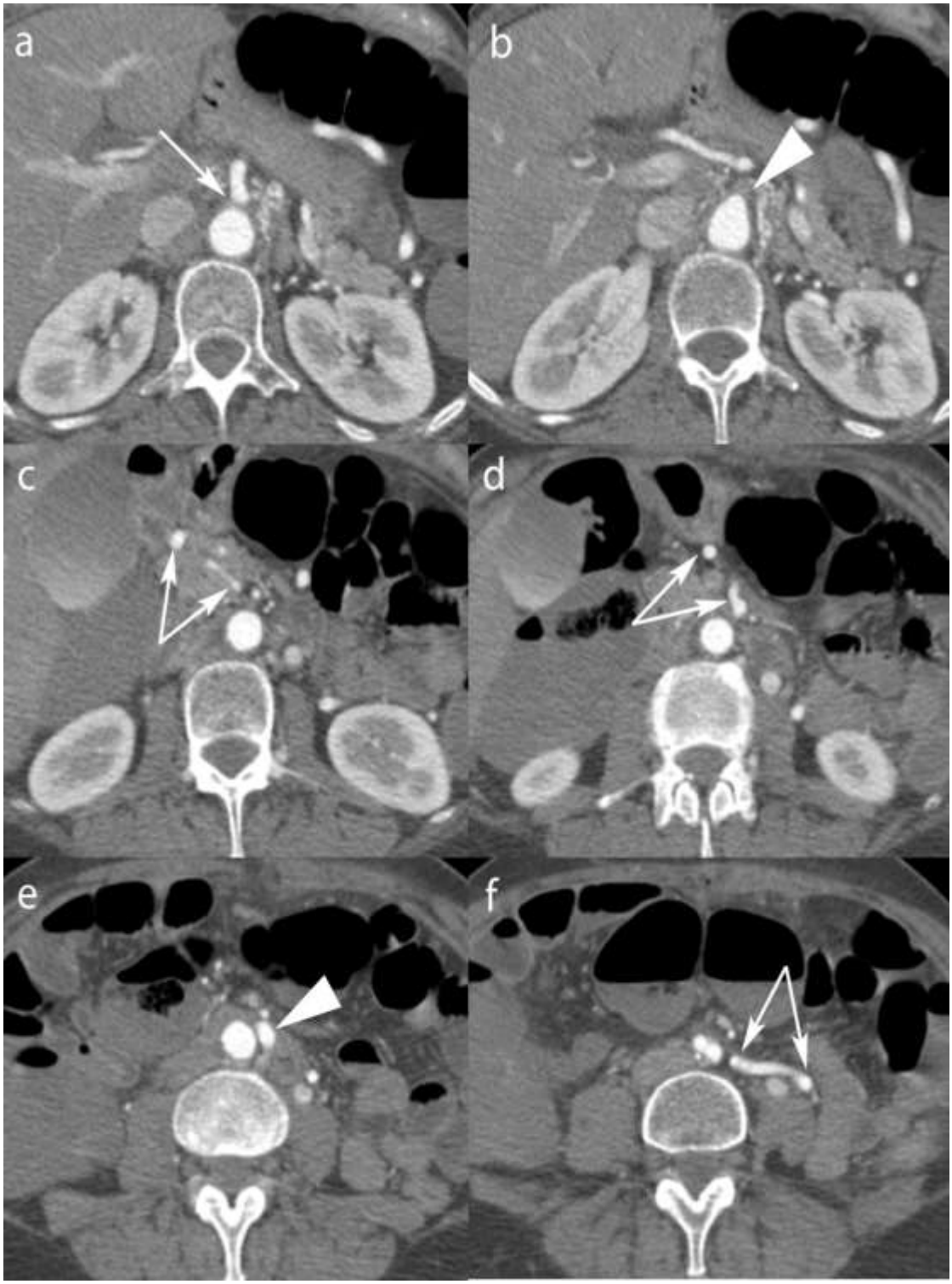
CASE REPORT

Chronic mesenteric ischemia (CMI) is a rare condition and which diagnosis may be delayed or missed as symptoms are unspecific before the occurrence of the classical post-prandial abdominal angina with significant weight loss.

The patient gave her written consent to the publication of these data.

The patient is a 59-year-old woman referred for chronic post prandial abdominal pain and weight loss (about 20 kg in the previous 6 months; BMI:16 kg/m^2). Her previous medical history was limited to hypertension and intestinal resection due to Meckel diverticula. She had an extensive gastrointestinal check-up in another hospital. A computed tomography angiogram (CTA) of the chest, abdomen and pelvis with multiplanar and centerline reconstructions of the mesenteric vessels revealed severe stenosis of the celiac artery, superior mesenteric artery (SMA) and inferior mesenteric artery (IMA) at their origin (Figure 1). The hypothesis of an endovascular repair of SMA was excluded, because the artery was occluded at the origin and for the first part and considered at high risk of complications. The patient received feedback about the diagnosis of CMI and a surgical revascularization was the recommended intervention.

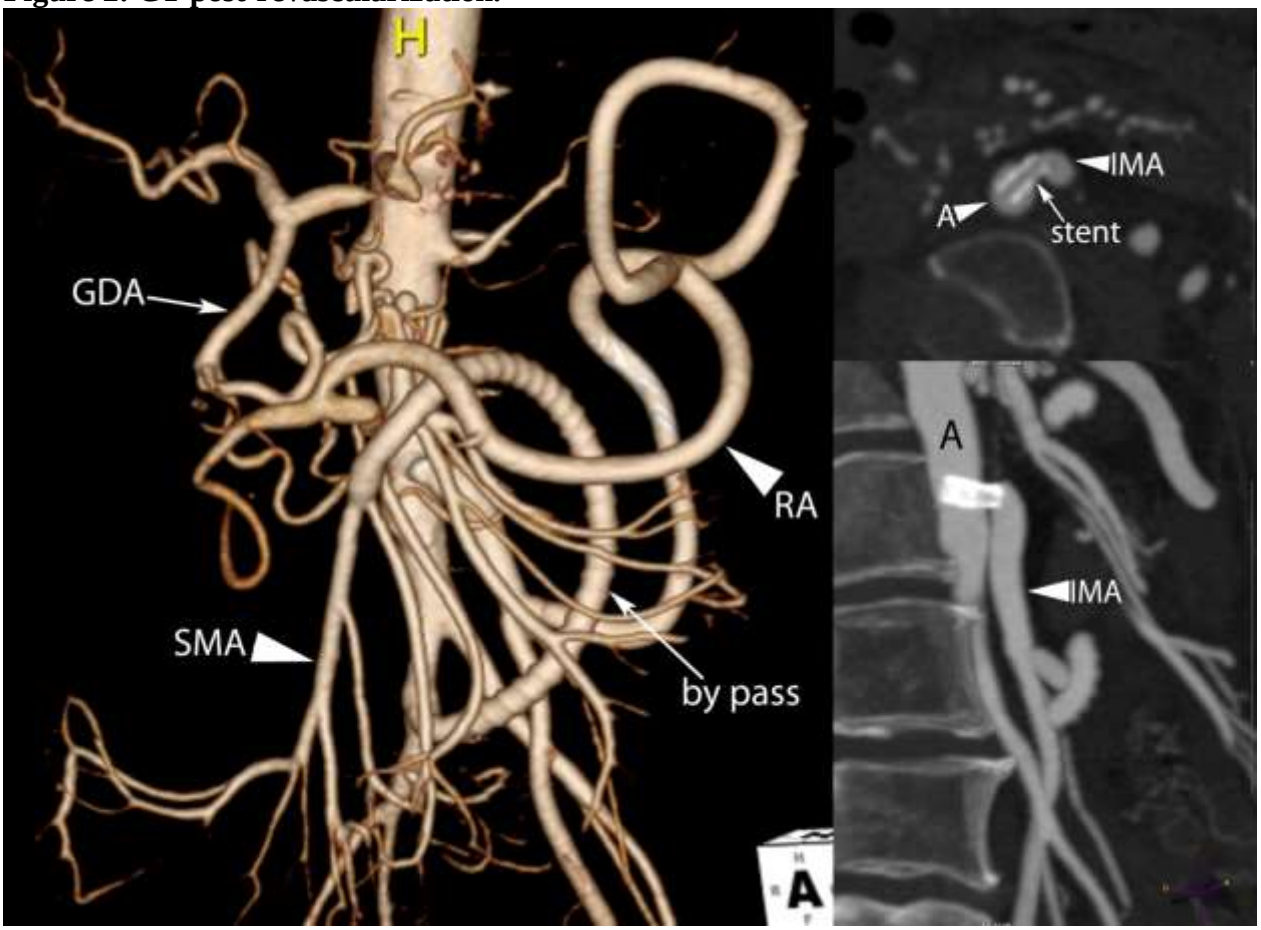
Figure 1: CT pre-revascularization.



Descriptive legend - a): ostial occlusion of celiac trunk; b): ostial occlusion of superior mesenteric artery; c): hypertrophic gastroduodenal artery (right) and pancreaticoduodenal artery (left); d): origin of hypertrofic Riolano arcade from superior mesenteric artery; e): post-stenotic dilatation of inferior mesenteric artery; f): connection between hypertrofic Riolano arcade and inferior mesenteric artery.

Through a median laparotomy, paravisceral aortic exposure was obtained. The SMA and the right common iliac artery were exposed. Subsequently, a retrograde iliac-SMA bypass was performed using a 6-mm diameter PTFE graft end-to-side with SMA. A 5x12mm balloon-expandable stent was inserted into the main trunk of the IMA, via retrograde IMA access (Figure 2). It was decided to treat IMA because intraoperative it appeared hypertrophic distal to the stenosis, that was significant, and the procedure was at low-risk of complications. The post-operative angiography showed that both procedures were successful.

Figure 2: CT post-revascularization.



Descriptive legend – GDA: gastroduodenal artery; SMA: superior mesenteric artery; RA: riolano arcade; IMA: inferior mesenteric artery; A: aorta; H: head.

Teicoplanine associated with ceftriaxone was introduced as the antibiotic therapy after the surgery. On the second post-operative day ceftriaxone was suspended, while meropenem was introduced and administered for 9 days. From the second post-operative day, the antiplatelet therapy was introduced consisting

in acetylsalicylic acid (100 mg die) and Clopidogrel (75 mg die).

On the second post-operative day, the patient developed chest pain associated with dyspnea. Electrocardiogram (ECG) showed sinus tachycardia. Troponin I was slightly increased (0,098 mg/dL). Her WBC counts increased to 19600/uL, platelet count was still within the normal range (196.000/uL). Creatinine was 0,5 mg/dL. Coagulation factors were in the normal range.

During the third post-operative day, dyspnea was persistent and the patient started to complain of abdominal pain, diarrhea, melena (3 evacuations per day) and she became olyguric. Laboratory tests revealed a persistence of neutrophilic leucocytosis with normal procalcitonin value, a remarkable anemia (haemoglobin dropped from 14,1 g/dL to 7,3 g/dL without any significant blood loss through the post-operative drainage), platelets count drop to 18.000/uL, elevation of serume creatinine to 1,85mg/dL, hyperbilirubinemia (total bilirubin 2,07 mg/dL, direct 0,94 mg/dL), and increase in transaminases (AST 82 U/L, ALT 40 U/L), aPTT, INR and fibrinogen levels were in the normal range. Blood smear examination revealed presence of schistocytes, indicating a thrombotic microangiopathy.

Transthoracic echocardiogram showed a FE>60%, normal inferior vena cava diameter and a possible subendocardial ischemia., Troponin I was unchanged. Chest X-ray was negative. CVP was 3 mmHg. A CT scan was performed and it showed a normal perfusion of the aortic-SMA by-pass and a normal perfusion of the IMA stenting, but colonic mucosa showed a clear thickening of the colonic wall.

During the night of the third post-operative day, the patient was treated with high volume fresh plasma infusion (30 ml/ kg bw) and 4 red blood cells units.

On the fourth post-operative day, the patient showed clinical improvements, and haemoglobin and platelets count reached the normal range. Troponine I level decreased. Fresh plasma infusion was repeated daily until the fifth post-operative day, when abdominal pain and diarrhea were no longer reported by the patient. Respiratory and renal functions improved and they became normal.

A multidisciplinary team composed by a nephrolgist, an immunologist, a hematologist and an interventional radiologist reviewed the case record and agreed that the etiopathogenesis was probably a thrombotic microangiopathy due to revascularization of both SMA and IMA. A CT scan performed during the 10th post-operative day showed that the by-pass and the IMA stent patency were associated with normal bowel perfusion. The patient was discharged on the 12th post-operative day.

A 3-month follow-up CT scan showed a regular bowel perfusion. The patient was asymptomatic and in good health. After about 10 months, her diet was unrestrained and she gained 7 kilograms in weight.

DISCUSSION

Chronic mesenteric ischemia (CMI) is an uncommon but severe disease in vascular surgery practice. usually caused by stenosis or occlusion of two visceral arteries. If untreated, it may cause death secondary to starvation or bowel infarction. There are multiple etiologies of it, the most common cause is atherosclerosis, and the other etiologies include fibromuscular dysplasia, aortic dissection, vasculitis, radiation, and cocaine abuse. Classically, the symptoms of CMI include abdominal angina which present as postprandial pain so as to make the patients develop a fear to eat, and at last lead to significant weight loss. But sometimes the symptoms are unspecific so this condition can be diagnosed late or misdiagnosed. Duplex ultrasound or CT angiography or MR angiography is available to reach the diagnosis. Open surgical or endovascular revascularization are two valid options in CMI (1-5), even if there is not an evidence-based technique recommended for its intervention. Morbidity and 30-day mortality differ based on the option chosen, but no cases of revascularization syndrome after treatment for/intervention of CMI are reported in literature.

In vascular surgery, Revascularization Syndrome (RVS) is a well-known complication, after a successful deep limb ischemia treatment and, even if with different patterns and prognosis, in cerebrovascular and spinal territories. The main characteristic of RVS is the cell damage, due to free radicals and ischemia's toxic products (6). Therefore, prognosis can be often lethal because of respiratory failure (ARDS), heart and renal failure (IRA) and hemorrhagic syndrome for the decrease of coagulation's factors.

Post-reperfusion syndrome after revascularization of mesenteric vessels is a very rare condition. A research in PubMed reported only one paper on the post-reperfusion syndrome, but it is referred to isolated intestinal transplantations (7). Regarding CMI, no cases are reported so far: the most recent literature studies reported data of morbidity and mortality when comparing different surgical approaches and endovascular treatments of CMI, but they do not mention the revascularization syndrome in patients treated with both procedures (1-5).

The case here illustrated is peculiar, given the very early onset of symptoms, with a fast occurrence of respiratory, hepatic and renal failure after the surgery. A sub-endocardial

ischemia was observed and the clinical evolution was complicated by hemolytic anemia and low level of platelets. The first hypothesis considered by the multidisciplinary team was a "thrombotic storm" due to a microangiopathy resulting in endothelial damage. Differential diagnosis has to take into account the group of the so called “thrombotic microangiopathy syndromes”, that share with CMI some common clinical and pathological features, including microangiopathic hemolytic anemia, thrombocytopenia and organ injury, as well as vascular damage. This is reflected in the pathological features such as abnormalities in the endotelium and vessel wall and arteriolar and capillary thrombosis (8).

The mucosal vascular alterations, due to reperfusion in this case, could have determined an endothelial damage playing an important role in the "thrombotic and inflammatory storm", as in catastrophic antiphospholipid syndrome (CAPS), a disorder similar to the 'classic' microangiopathic disorders, characterized by multiple simultaneous occlusions at a microvascular level (9,10). The pathogenetic hypothesis for this rare and severe variant of the classic antiphoshpholipid syndrome could have some similar aspects to the aforementioned case. Indeed, microangiopathic microvascular occlusions are themselves responsible for the ongoing thrombosis: clots continue to generate thrombin, fibrinolysis is depressed by an increase in plasminogen activator type-1, while the natural anticoagulant proteins, such as protein C and thrombin, are consumed. Pro-inflammatory cytokines, several products of the activated complement system, and antiphospholipid antibodies in the case of CAPS, are able to activate endothelial cells and up-regulate adhesion molecules and tissue factor (10).

The mucosal endothelial damage is the main physio-pathological mechanism hypothesized that can be complicated by "bacterial translocation", as it has been described in animal model (7). Indeed, the discontinuity of the mucosal barrier after ischemia/reperfusion injury can play a crucial role in determining the translocation of the enteric microorganisms from the gut to the bloodstream and to distant organs. Initial systemic inflammatory response syndrome (SIRS) may evolve in a septic condition.

The availability of a multidisciplinary consultation and an adequate resuscitation support with fresh plasma infusion, red cells transfusion and antibiotic therapy were crucial to the fast recovery and regularization of the anemia, platelet count, cardiac markers and renal respiratory functions (6).

CONCLUSION

This case illustrates a potential dramatic complication after revascularization of SMA and IMA in a patient with chronic mesenteric ischemia. For those patients undergoing an open surgical or endovascular revascularization for CMI, an early diagnosis should be considered, together with an aggressive treatment in the case of occurrence of a systemic inflammatory response syndrome.

We therefore suggest to treat these patients only in centers which have the availability of multidisciplinary monitoring and adequate treatment options.

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