Archives of the Balkan Medical Union
vol. 53, no. 2, pp. 293-296
June 2018

CASE REPORT

ACUTE ISCHEMIC CHOLECYSTITIS IN TAKAYASU'S SYNDROME – A RARE FINDING

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Received 17 Feb 2018, Accepted 30 Mar 2018

Abstract

Introduction. Takayasu's arteritis or pulseless disease is a systemic inflammatory disease of an unknown etiology, affecting medium and large arteries and their branches, leading to stenosis, occlusions, or aneurysmal degeneration. It is more frequent in young Asian women.

Case presentation. We present a rare case of Takayasu's disease in a young woman, who initially developed an acute ischemic cholecystitis. Ischemia is one of the mechanisms involved in the pathogenesis of acute acalculous cholecystitis. As this mechanism is most often involved in elderly people, such an uncommon finding at young age should be a reason to suspect a vasculitis with small vessel occlusion.

Résumé

Cholécystite acalculeuse et artérite de Takayasu – une découverte rare

Introduction. L'artérite de Takayasu ou la maladie sans pouls est une maladie inflammatoire systémique d'étiologie inconnue, affectant les artères moyennes et grandes et leurs branches, conduisant à une sténose, des occlusions ou une dégénérescence anévrysmale. Elle est plus fréquente chez les jeunes femmes asiatiques.

Rapport du cas. Nous présentons un cas rare de maladie de Takayasu chez une jeune femme qui a initialement développé une cholécystite ischémique aiguë. L'ischémie est l'un des mécanismes impliqués dans

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Conclusions. Acute cholecystitis in this patient was the first onset sign of Takayasu's arteritis.
An acute acalculous cholecystitis at young age could be of ischemic origin and should also be considered as a possible epiphenomenon of a generalized vasculitis disease.

Keywords: acute ischemic cholecystitis, acalculous cholecystitis, Takayasu's arteritis.

la pathogenèse de la cholécystite aiguë acalculeuse. Comme ce mécanisme est le plus souvent rencontré chez les personnes âgées, une découverte pareille à l'âge jeune serait un motif de soupçonner une vascularité avec occlusion des petits vaisseaux.
Conclusions. La cholécystite aiguë chez ce patient était un signe précoce de l'artérite de Takayasu. Une cholécystite acalculeuse aiguë à un jeune âge pourrait être d'origine ischémique et devrait également être considérée comme un épiphénomène possible d'une maladie vasculaire généralisée.

Mots-clés: cholécystite aiguë ischémique, cholécystite acalculeuse, artérite de Takayasu.

Introduction

Takayasu's arteritis or pulseless disease is a systemic inflammatory disease of an unknown etiology, involving medium and large arteries and their branches, leading to stenosis, occlusions, or aneurysmal degeneration. It is more frequent in young Asian women.

Ischemia is cited as one of the possible mechanisms involved in the pathogenesis of acute acalculous cholecystitis, proved by arteriography of gallbladder specimen. Small vessel occlusion, on the basis of low splanchnic flow or intravascular coagulation, may be a fundamental element in the pathogenesis of acute acalculous cholecystitis[1].

We found no literature report of association of Takayasu's syndrome with acute cholecystitis, only an atypical presentation with abdominal refractory pain[2].

Case report

We present the case of a 28-year old woman, of gipsy ethnia, who presented to our emergency room in November 2012 with symptoms of acute cholecystitis: pain in the right upper quadrant, vomiting and nausea. The symptoms progressed over the last 72 hours prior to presenting to the emergency room. The clinical exam revealed a rigid abdomen, tender to palpation in the right upper quadrant, fever 38°, pale skin and tachycardia (100 bpm). Laboratory results showed high white cell count (white blood cells 16.900/uL), neutrophilia (79.3%) and a high total bilirubinemia (2.30 mg/dL). We considered this high value as a severity marker in the context of acute inflammation, similarly with previously cited in cases of acute appendicitis, since no obstruction of common bile duct was demonstrated[3].

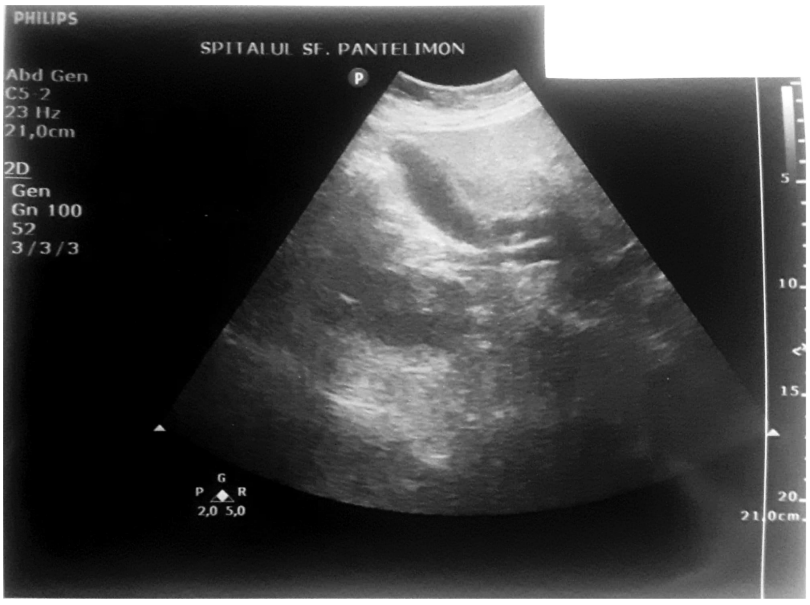

Figure 1.Ultrasonographic aspect of acute acalculous cholecystitis.

Abdominal ultrasound revealed thick (4-5 mm) and double contour gallbladder walls, low quantity of pericholecystic fluid (Figure 1) and plain abdominal radiography was without any anomalies.

The patient related a history of meningitis, probably of tuberculosis origin, with a decreased visual acuity one month before. She was admitted and started treatment with i.v. large spectrum antibiotics, nonsteroidal anti-inflammatory drugs and hydration.

After 24 hours, we performed a laparoscopic retrograde cholecystectomy under general anesthesia, with orotracheal intubation for acute acalculous cholecystitis.

At that moment, we only noticed the rare case of an acalculous acute cholecystitis in a young woman, an unusual event. The diagnosis of ischemic cholecystitis was confirmed by the histopathology exam that showed necrotizing ulcerative cholecystitis, with multiple arterial occlusions, with absent or minimal venous filling, in totally contrast with acute gallstone-associated cholecystitis findings, which usually exhibit arterial dilatation and extensive venous filling[1].

The postoperative outcome was good, the patient was discharged 48 hours after the surgical procedure, with the indication of coming back after one week for stitch removal.

The medical history became more and more complicated after that episode. In July 2013, she was admitted to an ophthalmology clinic, diagnosed with bilateral uveitis and retinitis and treated with corticosteroids. We could interpret today that episode as another epiphenomenon of Takayasu's vasculitis, as it is cited a similar case in the literature[4].

From April to October 2015, the patient was diagnosed with spastic paraparesis, tuberculous or viral meningitis and myelitis.

In August 2016, an abdominal ultrasound revealed a dilated abdominal aorta with thickening of its walls and a computed angio-tomography confirmed these findings and also showed narrowing of celiac brunch, occlusion of superior mesenteric artery, stenosis of renal arteries and severe distal abdominal aortic stenosis. The supposition of Takayasu arteritis was first established.

Between 18th of October 2016 and 3rd of March 2017 the patient was hospitalized in Torino, Italy. Antinuclear antibodies (anti DNA), ENA profile, ANCA and LAC were negative, temporal artery biopsy was negative, but periaortic biopsy confirmed the diagnosis of Takayasu arteritis. Bilateral renal arteries, celiac brunch and bilateral iliac arteries were stented. A restenosis of celiac brunch stent, with bowel occlusion and abdominal pain, imposed a repositioning of this stent.

In evolution, an aortic aneurysm with high rupture risk developed and imposed a third surgical intervention for correction, immediately after rescue treatment with high dose immunoglobulin (400 mg/kg, 4 consecutive days), associated to tocilizumab 8mg/kg single initial dose and corticotherapy 50 mg/kg. The spine MRI showed no vascular injuries of the spine, so the neural problems were recorded as a progressive meningomyelitis with spastic paraparesis. The coronarography exam was normal, despite a diffuse hypokinesia. No Koch bacillus identification technique was positive.

Nowadays, the patient is in a stable condition, under double antiplatelet therapy (aspirin, clopidogrel), following a rehabilitation physiotherapy program.

Discussion

Takayasu's arteritis was difficult to diagnose in this case. The diagnosis was established only after several years, during which the patient had several complications of this disease, interpreted as distinct other diseases.

Ischemic necrosis of the gallbladder is rare. Reported causes include acute bacterial endocarditis[5], rheumatoid arteritis[6], myocardial infarction[7], angiography[8], and visceral artery insufficiency[9], embolization of the cystic artery during hepatic arterial embolization for hepatic malignancies[10].

The gypsy ethnics in Europe, having been proved to come from India (Asia) people, seem to have a similar genetic predisposition for Takayasu.

A bilateral renal artery stenosis developed in this case, with secondary hypertension, resistant to treatment, similar with those in hemodialyzed patients, with uremic arteriopathy[11,12]. The differential diagnosis of renal failure in a young patient should be made with native kidney fibrosis, by ultrasound elastography[13].

Conclusions

Acute cholecystitis in this patient was the first onset sign of Takayasu's arteritis. An acute acalculous cholecystitis at young age could be of ischemic origin and should also be considered as a possible epiphenomenon of a generalized vasculitis disease.

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 all the patients included in the study"

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