

Ruptured mycotic hepatic artery aneurysm associated with methicillin sensitive staphylococcal endocarditis

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MVR and AVR operations were performed in Afshar Heart Center, Yazd, Iran and coil embolization was done in Cardiology Department, Shahid Sadoughi Hospital, Yazd, Iran

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Abstract

Hemobilia is a rare cause of upper gastrointestinal hemorrhage. Classical presentations of hemobilia are biliary colic, jaundice, hematochezia and melena. Persistent bleeding sometimes requires urgent therapeutic intervention, such as angiographic intervention or surgery. It may be secondary to trauma, inflammatory disorders, gallstones or tumors. Hepatic artery aneurysm rupture is a rare cause of gastrointestinal bleeding. Mycotic hepatic artery aneurysm is not a common vascular lesion, but it is considered to be a late complication of bacterial endocarditis. We present a 43 years old man with history of bacterial endocarditis that led to aortic valve replacement (AVR) and mitral valve replacement (MVR) about 9 months ago, with present complaint of rectal bleeding and dizziness. Angiographic studies suggested ruptured mycotic aneurysm in right hepatic artery, which was successfully treated with coil embolization.

Keywords

Hemobilia, Hepatic, Mycotic, Aneurysm, Bacterial endocarditis, Aortic valve replacement, Mitral valve replacement, Coil embolization

Imprint

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Introduction

The term hemobilia describes bleeding into the biliary system from any suggested origin [1]. One cause of hemobilia is rupture of hepatic artery aneurysm that is a rare vascular lesion [2] and can occur due to laparoscopic cholecystectomy, per cutaneous liver biopsy and blunt trauma [3]. Mycotic hepatic artery aneurysm is a vascular pathology which can be associated with bacterial endocarditis [4]. Its incidence is lowered due to development of effective antibiotics [5]. But if ruptures of these aneurysms occur, they are usually followed by fatal outcomes [6]. We present a patient with hemobilia due to the ruptured mycotic hepatic artery aneurysm as a result of Methicillin Sensitive *Staphylococcus aureus* (MSSA) bacterial endocarditis.

Case

A 43 years old man was admitted in emergency ward with neurological complaint such as cervical pain and upper limb paresthesia. The patient was febrile and during diagnostic work-ups Methicillin Sensitive *Staphylococcus aureus* (MSSA) endocarditis was suggested and echocardiographic studies revealed large vegetation on anterior mitral valve with severe mitral regurgitation and large vegetation on aortic valve with moderate to severe aortic insufficiency. According to acquired clinical and imaging data, septic emboli to cervical spinal cord and abscess formation was diagnosed to be the cause of his symptoms and thus effective antibiotics including Vancomycin 500mg QID, Gentamicin and ciprofloxacin 500mg BID were prescribed for him. Considering evidences of cervical cord compression, he was candidate for decompression surgery, but because of unstable hemodynamics and acute pulmonary edema due to severe destruction of two cardiac valves, he was transferred to cardiac operating room emergently to undergo aortic and mitral valve replacement surgery. The patient's neurological condition enhanced without any CNS intervention, and so antibiotic therapy was continued for him for 6 weeks, and the patient was discharged after an uneventful course in hospital under treatment of antibiotics and therapeutic dose of warfarin.

After 9 months, he was readmitted to hospital with epigastric pain, dizziness and rectal bleeding. In physical examination he was not febrile. His blood pressure was 80/60 mmHg, and his pulse rate was 114



Figure 1. Gastroduodenal artery and left hepatic artery normally branched from common hepatic artery.



Figure 2. Right hepatic artery branched from superior mesenteric artery instead of common hepatic artery as a normal variation

beats per minute; his conjunctiva was pale, and his sclera was icteric. Prosthetic aortic and mitral valve metallic sounds were auscultable, and epigastric tenderness was recorded. He was under treatment with warfarin 2.5mg daily. Laboratory data revealed the following data: white blood cell (WBC): 19800, hemoglobin (Hb): 6.2, mean corpuscular volume (MCV): 80, platelet (Plt): 129000, prothrombin time (PT): 18, partial thromboplastin time (PTT): 42 international normalized ratio (INR): 4/4, aspartate aminotransferase (AST): 561, alanine transaminase (ALT): 290,

alkaline phosphatase (ALP): 488, Total bilirubin: 2.5 and Direct bilirubin :1.4, blood urea nitrogen (BUN): 45 and creatinine (Cr): 0.9. Chest X ray findings were as follows: no acute lung process; the normal electrocardiogram (EKG) was recorded. He received two packed blood cells in emergency ward and underwent urgent upper gastrointestinal (GI) endoscopy. Endoscopic studies couldn't indicate any active bleeding site. In his abdominal ultrasound imaging the liver span was reported 107mm with heterogeneous and coarse parenchyma, dilated intrahepatic biliary ducts and sludge in gallbladder. No other pathology was reported in ultrasound imaging. The next day of hospitalization patient's condition regressed, and he complained of massive hematochezia. In physical examination he was still pale and icteric. His blood pressure was measured 90/40 mmHg, and his pulse rate was 106 beats per minute. Epigastric tenderness was recorded in physical examination. Measured Hb equaled 6.4. Urgent upper GI endoscopy was done for the second time. In second endoscopy there was no bleeding lesion along gastric mucosa, but there was fresh blood in second and third portion of duodenum without active bleeding site. Thus warfarin intoxication and bleeding from liver lesion or vascular pathology was suggested. Endoscopic retrograde cholangio-pancreatography (ERCP) investigation was suggested and done to find the bleeding source. ERCP results highly recommended diagnosis of hemobilia. The mentioned patient's past history of bacterial endocarditis with septic emboli to cervical spinal cord, infectious vascular lesion was highly recommended and considering patient's poor hemodynamic condition and low hemoglobin levels despite of receiving more packed cells in ICU, angiographic studies were indicated and done. In celiac artery angiography, the common hepatic artery, the gastroduodenal artery and the left hepatic artery were normal (Figure No.1), but the right hepatic artery was not visualized.

His superior mesenteric artery (SMA) was normal, but his right hepatic artery was branched from SMA as a normal variation (Figure No.2), and one aneurysmal lesion with active contrast leakage into biliary ducts was discovered (Figure No.3). Considering unstable clinical and hemodynamic condition, right hepatic artery embolization was performed using coil embolization technique.

After angiographic intervention and embolization, his bleeding stopped and his hemodynamics became

stable the next day, and he was finally discharged with therapeutic range of INR (Figure No.4).

Discussion

The term hemobilia was first used by Sandblom to describe bleeding into biliary system after a sub capsular liver injury in 1948 [7], but this term is now used to describe bleeding into the biliary system from any origin [1]. Bleeding caused by ruptured hepatic artery aneurysm (HAA) into the common bile duct was first reported by Jackson in 1821 [8]. In a major clinic study by Abbas and colleagues between 1980 and 1998, among 306 patients with visceral artery aneurysm diagnosis, the aneurysm was sited in HAA in 36 patients (12%). Among 36 patients with HAA aneurysm twenty-three patients (64%) were males, and 13 patients (36%) were females. The mean presentation age was 62.2 years (range, 20-85 year). HAA aneurysms are at definite risk of rupture (14%). Rupture risk factors include multiple HAA and non-atherosclerotic origin [9]. The most common sites of communication between the aneurysm and biliary tract are the common bile duct or hepatic ducts (76.2 per cent). Main features of hemobilia are biliary colic, jaundice, hematochezia and melena, and the bleeding may be recurrent. However, most patients do not present all four symptoms [10]. The pain originated from hepatic artery aneurysm is better sensed in the epigastrium or right upper quadrant or may radiate to the back [11]. The precise cause of HAA remains unclear yet. In the early twentieth century, most HAAs were mycotic, followed by bacterial endocarditis as the most frequent cause [5]. But nowadays mycotic HAAs are seldom seen, because of earlier antibiotic therapy for infections [12]. Mycotic hepatic artery aneurysm is important but is rare and accounts 0.1% of all arterial aneurysm [2]. At first, Osler described mycotic artery aneurysm in 1885 [13]. Its definition is an infectious break in artery wall that forms a blind-sac contiguous to arterial lumen. Maybe its pathogenesis is seeding of infected emboli into lumen of normal vessel leading to inflammation and necrosis and then weakening and dilation of arterial wall [14]. Most prevalent sites of involvement are the aorta, the peripheral artery, the cerebral artery, and the visceral arteries such as the superior mesenteric artery and the liver, spleen and kidney's arteries [15]. In hepatic artery aneurysm, the common hepatic artery is the most prevalent site (63%), and then the right hepatic artery (28%) is the most prevalent, but the left hepatic artery



Figure 3. Filling of right hepatic artery pseudo aneurysm with contrast before coil embolization.



Figure 4. Successful coil embolization.

(5%) is less involved, and at the end both right and left hepatic arteries involved together are the rarest cases (4%) [16]. Mycotic aneurysm in our patient was sited in his right hepatic artery, and mycotic hepatic artery aneurysms demonstrate high risk of rupture [4]. Most common etiologic pathogens are Streptococcus and Staphylococcus groups. Some other germs causing mycotic aneurysms are gram-negative bacteria, fungal infections such as aspergillus or candida, and even mycobacterium [15]. There are few cases of mycotic hepatic artery aneurysm with staphylococcal endocarditis in literatures [17, 18]. Staphylococci cause

20% to 30% of the cases of infectious endocarditis (IE), and 80% to 90% of these cases are caused by coagulase positive *Staphylococcus aureus* [19]. In a recent review of endocarditis cases at Duke University Medical Center from 1993 to 1999, in *S aureus* IE patients, 60% of the cases were involved by MSSA bacteremia, whereas 40% of the cases were involved by MRSA infection [20]. MSSA is associated with significantly higher rates of IE than MRSA. Community-acquired methicillin sensitive *staphylococcus aureus* (MSSA) is the cause of the most cases of MSSA endocarditis [21]. Hill and colleagues in 2008 reported that MSSA patients had significantly more unknown origin of bacteremia and experienced a significantly higher rate of major emboli compared to MRSA infected patients [22]. In our case, the patient was infected by methicillin sensitive *staphylococcus aureus*, which caused endocarditis with aortic and mitral valves involvement leading to AVR and MVR. Color Doppler ultrasonography, computed tomography (CT) scan, CT angiography and magnetic resonance (MR) angiography resulted in increased identification of asymptomatic hepatic artery aneurysms [23]. Magnetic resonance imaging can distinguish blood from stones and sludge and may be helpful when the diagnosis is uncertain. The diagnosis of hemobilia is most frequently confirmed by upper endoscopy or ERCP [24]. Today the most useful diagnostic test is selective arteriography of the celiac axis, the hepatic artery, or superior mesenteric artery [25]. Embolization with coil plays an important role in management of intrahepatic aneurysms in high risk surgery candidates and proximal aneurysm [9], thus for high risk surgery candidate patients arterial embolization is highly recommended, which is a simple, safe, and effective treatment for hemobilia with lower morbidity and mortality compared to surgery [26]. Open surgical treatment options for HAA include ligation, excision, venous grafting, synthetic grafting, and hepatic resection [5]. Surgical options are suitable for patients with low risk of surgery and usually distal aneurysms [27].

Statement on ethical issues

Research involving people and/or animals is in full compliance with current national and international ethical standards.

Conflict of interest

None declared.

Author contributions

All the authors read the ICMJE criteria for authorship and approved the final manuscript.

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