

# Hepatic artery aneurysm presenting with hemobilia in a patient with Behçet's disease: treatment with percutaneous transcatheteral embolization

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## ABSTRACT

We herein report a case of hemobilia caused by a hepatic artery aneurysm. A 40-year-old male patient with a history of Behçet's disease with a thrombus hanging out of the papilla of Vater had a large hepatic aneurysm in angiography. The hepatic artery feeding the aneurysm was embolized. Endovascular treatment can treat patients with hepatic artery aneurysms caused by Behçet's disease.

*Key words:* • Behçet syndrome • hepatic artery  
• aneurysm • embolization, therapeutic

Behçet's disease is a rare condition usually presenting with complex inflammatory manifestations like stomatitis, genital ulcerations, and iritis with sub-purulent products. The disease is common throughout the world, but is more prevalent in the eastern Mediterranean countries and Japan (1). The disease is of unknown origin with a histopathological picture revealing affected tissues that are infiltrated by mononuclear monocytes. The three above-mentioned main clinical manifestations, in combination with skin lesions like erythema nodosum, subcutaneous thrombophlebitis, and an acne-like exanthema, constitute the four major diagnostic criteria.

For the complete Behçet's syndrome to be diagnosed, all four major criteria must appear during the course of the disease (2). Two or three major criteria, including typical ocular infiltration, are called "the incomplete Behçet's syndrome" and the potential diagnosis is amplified by the presence of one or more minor criteria (2).

Rare symptoms and complications that may also be observed are arthritis (similar to the type associated with inflammatory bowel disease), enterocolitis, epididymitis, and involvement of the central nervous system.

Vascular inflammatory lesions, which can affect both arteries and veins, may lead to large aneurysms and vascular obstructions. These large aneurysms represent the most feared complications of the disease, because they can complicate a patient's situation and result in surgery. Endovascular treatment with arterial embolization is a less invasive therapeutic option. Herein, we present a patient with Behçet's disease, who had a rare hepatic artery aneurysm, which ruptured into the biliary system causing hemobilia. The aneurysm was successfully treated by endovascular embolization.

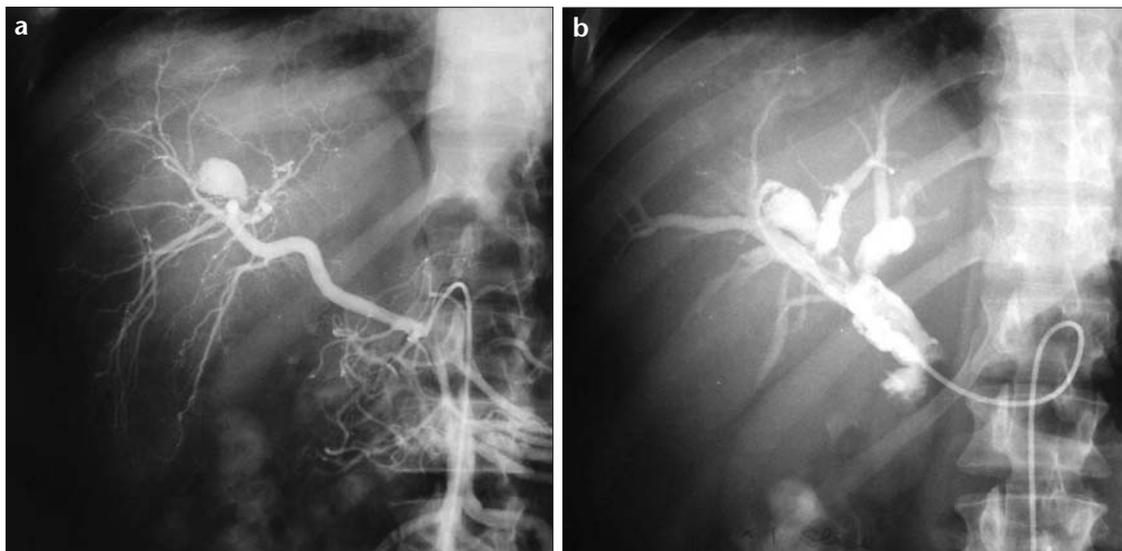
## Case report

A 40-year-old male patient with a history of Behçet's disease (initially presenting with iritis and genital ulceration; later with stomatitis, bilateral malleolar arthritis, and recurrent sciatic pain) was referred to us with upper right abdominal quadrant colic and vomiting. Prior to this referral, computed tomography (CT) of the abdomen had recently been performed due to recurrent episodes of epigastric discomfort and was normal. The patient had been treated with steroid agents for Behçet's disease between 1995 and 1999.

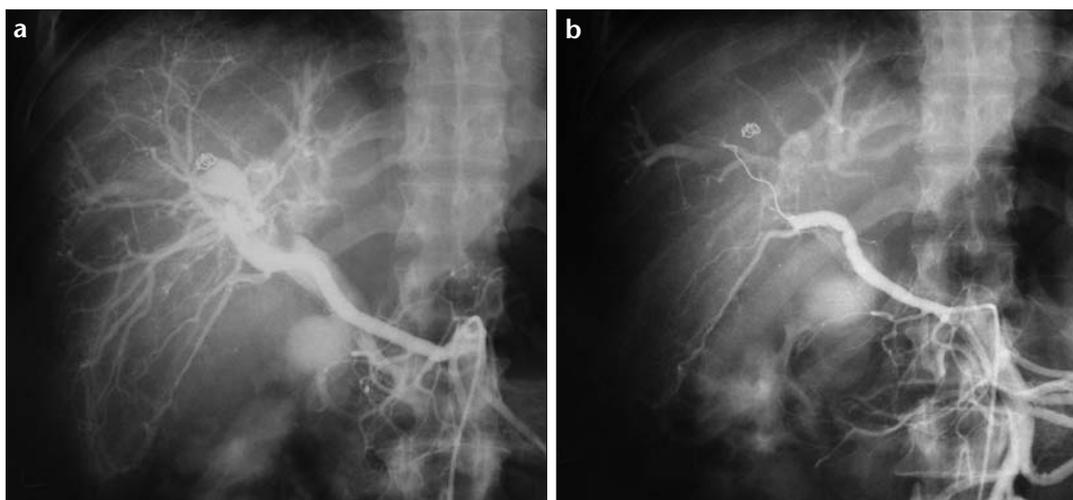
On his last admission, the patient presented with clinical and biochemical findings of acute pancreatitis. Ultrasonography (US) of the liver, pancreas, and biliary system revealed bile sludge in the gallbladder and findings of focal pancreatic head inflammation. Since the patient did not improve, endoscopic retrograde cholangiopancreatography was performed for common bile duct inspection. A thrombus protruding from the papilla of Vater was detected; therefore sphincterotomy

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**Figure 1. a, b.** Right hepatic artery opacification after catheterization of the superior mesenteric artery shows a large aneurysm in the right liver lobe (a). Superselective catheterization of the aneurysm's neck revealed extravasation of the contrast medium into the biliary system. The bile ducts are dilated due to large thrombi causing intraluminal-filling defects (b).



**Figure 2. a, b.** After placement of a 5x30 mm platinum coil, the coil fell into the aneurysm neck, so only partial thrombosis of its lumen was noted (a). A second platinum coil 4x30 mm was placed into the feeding artery. The coil diameter was too large for the artery, so it was not optimally curved and caused an extended thrombosis of the right hepatic artery. The aneurysm was successfully thrombosed as well (b).

was not attempted. After thrombus removal, uncontrollable hemorrhage occurred and the procedure was interrupted. The patient's hematocrit level decreased 15 units during the next 24 hours. Emergency angiography was performed and selective catheterization of the celiac trunk revealed a normal left hepatic artery. The right hepatic artery was found originating from the superior mesenteric artery with a large aneurysm lying intrahepatically (Figure 1a). Superselective catheterization of the aneurysm's neck showed massive extravasation into the biliary tract, which was dilated due to large intraluminal thrombi (Figure 1b). A platinum

coil 5 mm in diameter and 30 mm long (Cook, Bloomington, USA) was inserted in order to embolize the aneurysm's neck, but only partial thrombosis was achieved (Figure 2a). Subsequently, embolization of the feeding arterial branch was performed by introducing a coil of the same material and type, which was 4 mm in diameter and 30 mm long. Unfortunately, the coil was too wide for the vessel, so it did not optimally curve, causing thrombosis of the larger part of the right hepatic artery (Figure 2b). Nevertheless, the aneurysm thrombosed completely and hemobilia was terminated. For several hours, the patient experienced only moderate pain,

which was conservatively treated. Liver enzymes, which were mildly elevated, normalized after two weeks. Follow-up CT two months later revealed no infarction in the liver.

### Discussion

Behçet's disease is an uncommon systemic process that appears most often in the third or fourth decade of life (3). It is characterized by recurrent orogenital ulcers, ocular and cutaneous inflammatory lesions, as well as cardiovascular involvement with large aneurysm formation commonly in the pulmonary and carotid arteries, and also in peripheral and visceral vessels

(3, 4). Such vascular inflammatory lesions can affect medium size arteries, like the hepatic artery, leading to hemorrhage or hemobilia, as in our case. These complications may negatively affect a patient's condition, resulting in increased morbidity or mortality.

There is no specific treatment for Behçet's disease. Colchicine is used for the relief of skin ulceration and iritis and steroids for central nervous system and ocular involvement. In cases that are resistant to steroids, immunosuppressive agents such as azathioprine and cyclosporine can be used. Despite the low mortality rate of 3-4%, the prognosis is relatively poor (1, 2).

In cases that entail vascular aneurysms, surgical treatment is an option, with reconstruction of the diseased artery by a PTFE (polytetrafluoroethylene) graft (5). Nevertheless, such treatment is not always possible, and other therapeutic alternatives such as percutaneous transcatheter embolization, with or without balloon occlusion, can be applied (6, 7). Pulmonary arterial aneurysms causing hemoptysis can be percutaneously embolized, usually with good results (7). Successful surgical aneurysm repair in patients with Behçet's disease is reported in the literature (8), but liver involvement was not present in any of these cases.

Hemobilia, induced by percutaneous iatrogenic punctures, which cause pseudoaneurysms of the hepatic artery, is a well-known complication. Successful embolization with the use of stain-

less steel coils is described in the literature (9). Hepatic artery involvement of Behçet's disease treated by means of endovascular embolization, however, is quite rare in the literature. Endovascular treatment of a massive hepatic artery aneurysm, which caused arterioportal fistula with the superior mesenteric vein, has been described once (10). To the best of our knowledge, our case of an aneurysm causing hemobilia in a patient with Behçet's disease is the first to be reported. The therapeutic options for this patient were either an extended right hepatectomy or transcatheter embolization. The right hepatic artery occlusion was not our planned treatment technique, but it did not lead to any severe implications. There were two reasons for this: a) the anatomical variation and separate liver perfusion through the large left hepatic artery leading from the celiac trunk; b) adequate portal vein blood supply.

In conclusion, we believe that endovascular embolization of hepatic artery aneurysms in patients with Behçet's disease can be considered as a first-line treatment in emergency cases because it is a minimally invasive technique with a low rate of morbidity.

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