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BRIEF REPORT

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Life-threatening gastrointestinal bleeding after liver transplatation due to hepatic artery pseudoaneurysm perforating into the common bile duct. A case report

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Abstract A 54-year-old male presented with acute rejection and life-threatening gastrointestinal bleeding 2 months following orthotopic liver transplantation. Since no bleeding was identified in the entire gastrointestinal (GI) tract, hematobilia was first suspected and an arteriocholedochal fistula angiographically confirmed. Two days after resection of a pseudoaneurysm of the hepatic artery (HA) with primary repair and closure of the bile duct fistula, hepatic artery thrombosis (HAT) occurred. Various attempts to revascularize the HA eventually failed. Two weeks later, a CT scan showed necrotic areas within the two left lateral segments. At relaparotomy, major parts of the bile duct were found to be necrotic, and the biliary anastomosis was therefore abandoned and necrotic tissue removed. HAT was otherwise well tolerated by the graft and, at a further relaparotomy some weeks later, a hepaticojejunostomy was performed. Two years after transplantation the patient is well with a normally functioning graft.

Key words Liver transplantation, pseudoaneurysm hepatic artery · Hepatic artery pseudoaneurysm, liver transplantation · Hematobilia, liver transplatation

Introduction

Hepatic artery thrombosis (HAT) is a frequent complication of orthotopic liver transplantation, its incidence in adults ranging from 5 % to 10 %. [1, 3, 9] In some cases it has been possible to successfully treat HAT by surgical or pharmacological means. When this complication occurs during the early postoperative period and such therapeutic attempts fail, retransplantation is usually required [1, 4, 10]. However, HAT may be tolerated by the graft if it happens 2 or more months after transplantation [3, 10]. In contrast, pseudoaneurysm of the HA after liver transplantation is a much rarer complication, with fungal infection or technical problems being the main etiological factors [7, 11]. Such a pseudoaneurysm may perforate into the common bile duct and thus cause life-threatening hemorrhage. Only very few such cases have thus far been reported.

We report here on what is, to the best of our knowledge, the first patient to develop an arteriocholedochal fistula after rupture of a pseudoaneurysm with massive intestinal bleeding. Despite ligation of the HA and abandonment of the bile duct anastomosis, we were able to save the graft and eventually create a hepaticojejunostomy.

Case report

Orthotopic liver transplantation was performed on 10 April 1993 on a 54-year-old male suffering from end-stage liver cirrhosis of unknown etiology. Venous-venous bypass surgery was performed without any complications. For arterial reconstruction, donor common HA was anastomosed to the recipient common HA in an end-to-end fashion using magnifying glasses. Finally, an oblique choledochocholedochostomy was performed. The patient received a total of 13 units of red blood cells. Prophylactic immunosuppression

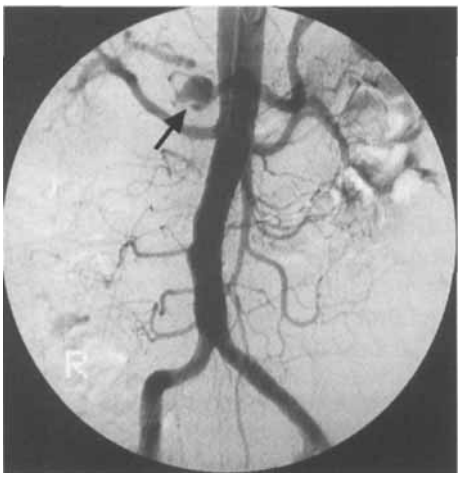

Fig. 1 Angiographic depiction of hepatic artery pseudoaneurysm

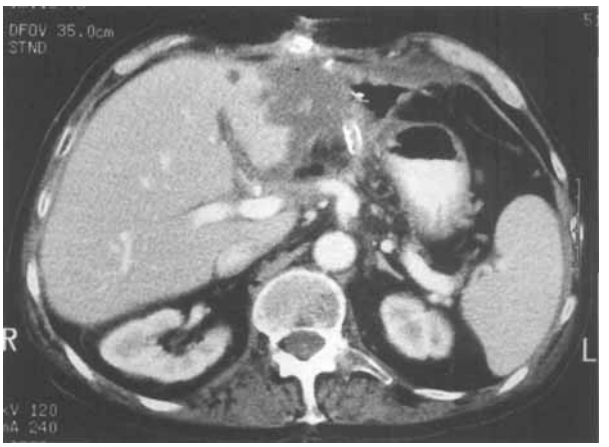

Fig. 4 CT scan showing necrotic areas within the left lateral segments of the liver

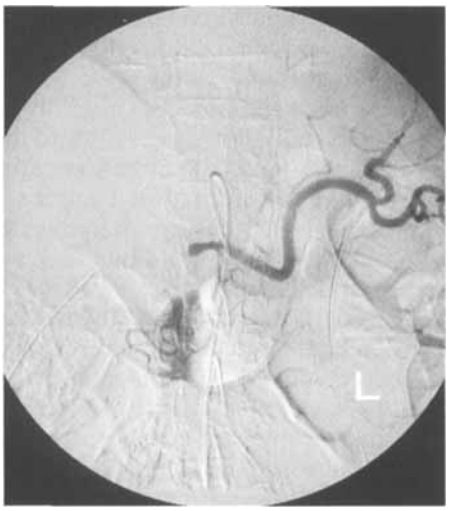

Fig. 2 Hepatic artery thrombosed

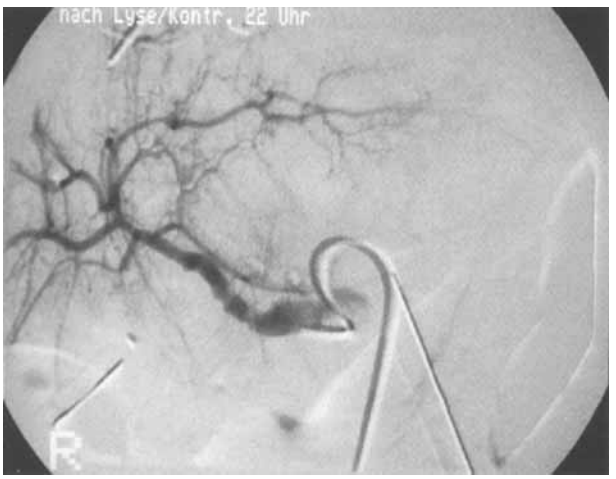

Fig. 3 Patency of hepatic artery after local thrombolysis

consisted of cyclosporin (CyA), steroids, and azathioprine. The postoperative course was completely uneventful and the patient was discharged on postoperative day 25 with normal liver function tests but slightly elevated creatinine values.

Two months after transplantation the patient presented with right upper abdominal pain and recurrent melena. During this period the patient became intermittently jaundiced and a drop in hemoglobin was noted. Elevated levels of liver function tests (total bilirubin 3.05 mg/dl; GOT 95 U/l; GPT 371 U/l) were thought to be due to rejection since CyA levels had fallen below 50 ng/ml and mechanical obstruction was ruled out by ultrasound. The clinical diagnosis was confirmed by graft histology. The patient was therefore given 500, 250, and 125 mg methylprednisolone on 3 consecutive days, and desired CyA blood levels were quickly reached by intravenous administration of the drug. All parameters returned to normal shortly thereafter. A few days later the patient again presented with melena and a further distinct drop in hemoglobin and hematocrit. Neither gastroscopy nor colonoscopy could reveal the source of the bleeding. No further GI bleeding occurred until some days later, when the patient had to be readmitted for hemorrhagic shock. Endoscopy again failed to localize the bleeding site. It was at that moment that bleeding from the HA into the bile duct was first suspected and angiographically confirmed. During this bleeding episode, liver function remained stable.

At laparotomy, a pseudoaneurysm of the donor HA was identified with an arteriocholedochal fistula at the site of the bile duct anastomosis but 3 cm distal of the arterial anastomosis (Fig. 1). Since the operating field did not seem to be contaminated, the pseudoaneurysm was resected, the HA reconstructed by direct anastomosis of the cut vessel ends, and the bile duct leak closed. However, enterococci and *Candida* were cultured from the specimen and the patient was therefore given piperacillin and ketokonazol.

Two days later, an increase in liver function parameters was again noted. Doppler ultrasound revealed arterial thrombosis. After angiographic confirmation, thrombectomy was performed and part of the donor HA resected and reconstructed using a saphenous venous autograft. Because of a prothrombin time greater than 120 s due to overheparinization, the patient was given two units of fresh frozen plasma. Arterial rethrombosis was identified by Doppler ultrasound only 1 day later. A catheter was then selectively inserted into the HA and lysis of the thrombus achieved by

local administration of regional tissue plasminogen activator (Figs. 2, 3). Immediate, postinterventional patency of the HA was demonstrated by Doppler ultrasound.

Two weeks later, a CT scan showed necrotic areas within segments II and III of the left lobe of the liver and a hematoma at the site of the upper caval anastomosis (Fig. 4). At relaparotomy, which was performed on 31 July 1993, the HA was found to be thrombosed. The necrotic areas were resected and the hematoma removed. The HA was ligated, the leaking biliary anastomosis abandoned, and a catheter inserted into the bile duct for temporary drainage to the exterior. At that time, the bile was found to be infected with *Candida albicans* and a multiresistant enterococcus faecium, which were successfully treated with amphotericin B and imipenem-cilastatin.

At a further laparotomy on 17 August 1993, some necrotic liver tissue was resected from the left liver lobe; the bile duct, however, was found to be vital. A hepaticojejunostomy (Roux-en-Y) was performed. No steroids were given during the first 2 postoperative weeks in order to protect the biliodigestive anastomosis. The further clinical course was uneventful and the patient was discharged on 5 September 1993. He continues to have normal liver function tests 2 years post-transplantation.

Discussion

Arterial pseudoaneurysm is a rare complication following orthotopic liver transplantation and usually occurs at the site of the arterial anastomosis. Infection is thought to be the most frequent cause, but needle biopsy and transhepatic catheter insertion have also been discussed as possible causes of this complication [7, 11]. Fistulas between the HA and the common bile duct or the upper GI tract are even less frequent. Thus far, only one patient with an arteriocholedochal and two patients with an arteriogastric or arterioenteric fistula have been described by the Pittsburgh group [7]. All of them had a pseudoaneurysm of the HA and presented with severe GI bleeding, which led to the diagnosis. In four of the five cases a biliary leak was thought to be the cause of pseudoaneurysm. All but one occurred between days 54 and 60, the remaining case on day 10. The HA was ligated in four cases and embolized in one. Two of the five patients survived with normal liver function, whereas two patients died from *Candida* peritonitis. The remaining patient, in whom the fistula occurred soon after transplantation, developed acute hepatic failure. Five days following retransplantation he succumbed to multiorgan failure and sepsis.

Our patient presented with a histologically confirmed rejection episode and recurrent GI bleeding without an endoscopically detectable source within the entire GI tract, which first made us think of an arteriocholedochal fistula [7, 11]. In retrospect, we believe the rejection episode was clearly related to the low CyA level triggered by hematobilia, which should be included in the differential diagnosis of a jaundiced patient after orthotopic liver transplantation. Interestingly, the pseudoaneurysm was found not at the site of the arterial anastomosis but three centimeters above. Although not proven, it could well have been at the origin of the gastroduodenal artery. Since no needle biopsy or transhepatic catheterization was performed in this patient, the bile leak must be considered the cause of pseudoaneurysm [7, 11]. The operative field did not look contaminated; thus, the pseudoaneurysm was resected, the direct repair performed, and the bile leak closed.

When arterial thrombosis occurred 2 days later, thrombectomy was performed, major parts of the HA resected, and a saphenous venous graft used for reconstruction. At that time, the result of the culture was still unknown. Thrombolytic treatment for rethrombosis was only temporarily successful [5]. Ligation of the HA thereafter resulted in small necrotic areas at the left lobe; HAT was otherwise well tolerated by the graft. This is not unusual when it happens 2 month after transplantation [2, 3, 10]. Sufficient arterial revascularization of the bile duct system must be assumed at the time of the hepaticojejunostomy [8].

Even if there is no macroscopic evidence of bacterial or fungal infection and other causes of pseudoaneurysm can be ruled out, contamination must be anticipated [11]. In this situation, arterial reconstruction, even without prosthetic material, is not successful and thus should probably be avoided. Surgical revascularization can be attempted if no bile leak is identified and if sterile conditions can be expected. In the Pittsburgh series, the coincidence of pseudoaneurysm and bile leak was associated with a high mortality (3/4 patients) [7].

In the presence of severe GI bleeding without an endoscopically detectable source or abdominal bleeding, a ruptured pseudoaneurysm of the hepatic artery must be considered in liver recipients [7, 11]. Diagnosis is easily confirmed by angiography but may be difficult or impossible by Doppler ultrasound [11]. In some cases ligation of the HA may save these patients, be well tolerated by the graft, and not even lead to bile duct necrosis when carried out 2 months or more following transplantation [3, 4, 6]. However, retransplantation may be required if it happens during the immediate postoperative period.

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