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CASE REPORT

Arterioportal Fistula Following Liver Biopsy Three Cases Occurring in Liver Transplant Recipients

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Liver biopsy is a diagnostic procedure which is utilized frequently in liver transplant recipients. Here we report the experience of the University of Pittsburgh with an unusual complication of this procedure occurring in liver transplant recipients.

CASE REPORT

Case 1. A 26-year-old woman underwent orthotopic liver transplantation (OLTx) for end-stage liver disease secondary to non-A, non-B hepatitis under cyclosporine and prednisone immunosuppression. She experienced acute cellular rejection on the fifth postoperative day documented by a percutaneous liver biopsy. She was treated with a steroid bolus and an increase in her baseline immunosuppression. A hepatic arteriogram obtained to evaluate the patency of the hepatic artery was performed two weeks after liver biopsy. It documented an incidental peripheral right hepatic artery to right portal vein fistula (Figure 1). The patient was asymptomatic without any evidence of portal hypertension and remained so for over five years. She was followed with repeated Doppler ultrasound studies of the liver obtained monthly for 3 consecutive months and then annually for over five years until the fistula closed spontaneously.

Case 2. A 64-year-old man with chronic active hepatitis underwent orthotopic liver transplantation under cyclosporine and prednisone immunosuppression. Three months after transplant he was admitted for elevated liver enzyme levels and was found to have a stricture at the choledochcholedochostomy site with a dilated intrahepatic biliary system. The biliary anastomosis was revised using a Roux-en-Y choledochojunostomy. An intraoperative liver biopsy was obtained from the left hepatic lobe. The subsequent postoperative course was complicated by intermittent gastrointestinal tract bleeding and hyperbilirubinemia. A Doppler ultrasound study of the liver was unremarkable. A

percutaneous transhepatic cholangiogram revealed complete biliary obstruction at the level of the biliary-enteric anastomosis. A second abdominal exploration revealed hemobilia with blood clot filling the entire biliary system. No active bleeding was evident. The biliary system was irrigated and the biliary anastomosis was revised. Postoperatively the patient experienced recurrent hemobilia. An angiogram was obtained and documented a left hepatic artery to left portal vein fistula. Embolization was performed with transient closure of the fistula. However, recurrent episodes of hemobilia documented by radionuclide scanning persisted. A repeat hepatic arteriogram showed persistence of the fistula in the left side of the liver. A second attempt at hepatic arterial embolization was attempted unsuccessfully. Ultimately, a second liver transplant was performed six weeks after the initial diagnosis of the arterioportal fistula. Examination of the resected allograft revealed multiple areas of infarcted liver, thrombosis of the main hepatic artery, and large colonies of gram-positive cocci within the necrotic liver tissue. The second liver failed as part of a syndrome of multiple organ failure and sepsis.

Case 3. A 17-month-old boy with cat's eye syndrome underwent orthotopic liver transplantation at the age of 11 months. He received the left lobe graft of a split liver. His postoperative course was complicated by an intraabdominal abscess and six episodes of rejection, each of which was confirmed by liver biopsy. All six liver biopsies were performed utilizing ultrasound guidance and using a Menghini needle. One of the biopsies was associated with bleeding at the percutaneous biopsy site. Three months later, the patient was readmitted because of mild elevations in the serum aminotransferase levels. His physical examination was unremarkable except for a bruit over his liver. He had no signs of portal hypertension or congestive heart failure. A seventh liver biopsy revealed lobular and portal inflammation as well as central venous intimal fibrosis. An hepatic arteriogram performed because of a suspicion of hepatic artery occlusion demonstrated a large arteriovenous fistula (Figure 2). The fistula was also subsequently identified with duplex ultrasound. Upper gastrointestinal endoscopy and echocardiography were normal. Three months later, following a response to an increase in the amount of immunosuppression being used, he had normal liver biochemistries. He has had no evidence of portal hypertension following the transplant in over three years of follow-up, although the fistula has not changed in size.

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Fig 1. An arterioportal fistula (arrow) in the peripheral right hepatic lobe is demonstrated. Note filling of small portal venous branches distal to the fistula.

DISCUSSION

The existence of an hepatic arterioportal fistula was first reported by Goodhart (1). Since then, a total of 282 cases of hepatic arterial to portal vein fistulas have been described (2). None, however, have been reported following a liver transplant. Acquired hepatic arterial portal venous fistulas can occur spontaneously due to the rupture of an arterial aneurysm into the portal system as a result of neoplastic growth or, more commonly, as a result of trauma to the liver (either blunt or penetrating) (3). Iatrogenic fistulas, occurring as a result of a liver biopsy percutaneous transhepatic cholangiogram, or transhepatic biliary catheterization, represent 60% of all published cases.

Since the first postliver biopsy arterioportal fistula was reported by Preger in 1967 (4), almost 100 additional cases have been reported in the English literature alone. Okuda et al reported an arterioportal fistula rate of 5.4% occurring in 93 patients who underwent hepatic arteriography after an earlier liver biopsy (5). Hellekant and Olint (8) reported a 52%

incidence of vascular abnormalities after liver biopsy if the subsequent arteriogram was performed within a week of the liver biopsy. Only a rate of 10% for vascular abnormalities was found if the arteriogram was delayed until three weeks after liver biopsy (6). In a multicenter retrospective study of 68,276 liver biopsies performed in Europe, no hepatic arterioportal fistula were found (7). Such a discrepancy in case reporting can be attributed, at least in part, to the asymptomatic nature of most hepatic arterial portal venous fistulas and their tendency to close spontaneously (5, 8, 9). Occasionally, an arterioportal fistula of the liver can cause portal hypertension, which can become clinically manifest as either the presence of ascites or esophageal varices (10, 11). In contrast to systemic arteriovenous fistulas, which can cause congestive heart failure, such has never been reported to occur with an iatrogenic arterioportal fistula.

The management of hepatic arterioportal fistulas, once detected, is controversial. If there is evidence of portal hypertension, closure of the fistula is essential.



Fig 2. Very large arterioportal fistula in the left hepatic lobe of a split liver graft is demonstrated on a selective hepatic arteriogram. Note the markedly enlarged hepatic artery.

The preferred method for intrahepatic fistulas is percutaneous embolization (12, 13). Surgery is an option for extrahepatic fistulas, if hepatic arterial embolization therapy fails to close the fistula.

Asymptomatic fistulas need not be treated. Their clinical course can be followed using serial duplex ultrasound studies. Treatment of asymptomatic fistula should be considered if an increase in size or signs of portal hypertension develop.

Case 1 demonstrates a typical case of an asymptomatic fistula that closed spontaneously. In contrast, case 2 represents a case complicated by recurrent severe hemobilia, which eventually led to graft failure and death. The fistula in this second case most likely developed after an intraoperative liver biopsy performed at the time of the original transplant, which produced both an hepatic arterial biliary fistula and an hepatic artery portal vein fistula. Although embolization of an hepatic arterioportal fistula is the preferred treatment for symptomatic cases, it is not always successful, as was the situation in this more

complex case with both an hepatic artery biliary fistula and an hepatic artery portal vein fistula. An early left lateral segmentectomy may have been a better therapeutic choice in this case but was not possible later when multiple hepatic infarcts had developed as a result of the earlier attempts of hepatic arterial embolization. In the third case, the arterioportal fistula was most likely the result of a percutaneous liver biopsy, although the possibility that this complication occurred as a result of splitting the donor liver must at least be considered. A normal duplex scan obtained in the first posttransplant month, a history of a complicated percutaneous liver biopsy, and the subsequent development of a bruit, all suggest that the fistula developed as a result of a liver biopsy. The treatment options in this third case were limited because of the proximity of the fistula to the main hepatic arterial trunk. In this particular pediatric patient, a selective hepatic arterial embolization was thought to be too dangerous because of the risk of a thrombosis of the hepatic artery. Moreover, because surgical interven-

tion also had risks, it was elected to follow the fistula longitudinally. The patient has been followed on a regular semiannual basis with serial duplex scans. No change in the fistula has occurred.

SUMMARY

Although liver biopsy is a very useful procedure used frequently in the diagnosis and management of liver dysfunction occurring after orthotopic liver transplantation, complications can occur with its use. An unusual complication of arterioportal fistula is reported here.

Based upon this small series of an unusual event and the knowledge that the posttransplant liver may be more hypervascular than prior to OLTx and that it is uniquely susceptible to hepatic infarction and abscess formation, any attempt at fistula closure should be considered carefully prior to initiating the therapy (15). Unless a serious complication occurs [such as a transient biliary obstruction due to hemobilia as occurred in case 2, portal hypertension as also occurred in case 2, or systemic sepsis or other symptoms develop related directly to the fistula], simple observation may be the best choice of action. Should therapy be required, hepatic arterial embolization should be reserved for adults with intrahepatic fistulas. Primary surgical closure of intrahepatic fistula should be reserved for cases of extrahepatic fistula.

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