

## A Case Report of Hepatic Artery-portal Vein Fistula with Portal Hypertension

Tosiomi KUSANO, Noboru HARADA, Nobuyuki MINAMI,  
Yoshiteru KUSANO, Ryoichi TSUCHIYA

*The Second Department of Surgery,  
Nagasaki University School of Medicine,  
Nagasaki, Japan*

Received for publication, June 30, 1985

A rare case of portal hypertension secondary to traumatic hepatic artery portal vein arteriovenous fistula (A-P fistula) due to liver needle biopsy was reported. Successful resection of the A-P fistula in this patient was made by a limited partial hepatectomy under guidance of intraoperative ultrasonic evaluation of the site of the fistula. In high risk cirrhotic patient, mass resection of the liver parenchyma is usually fatal. Therefore, the reported cases in the literature were treated by conservative such as ligation of the artery, and the result was almost not acceptable. In this report, we emphasized the usefulness of intraoperative orientation of the A-P fistula by ultrasonic examination which allowed a limited partial hepatectomy for complete removal of the affected area without any postoperative complication.

### INTRODUCTION

Since SACKS<sup>7)</sup> reported a case of hepatic artery-portal vein arteriovenous fistula (A-P fistula) who died of bleeding from esophageal varices due to portal hypertension in 1982, 47 patients with hepatic A-P fistula have been appeared in the literature. In Japan, there have been only a few cases such as by OKUDA<sup>5)</sup>, and HARADA<sup>3)</sup> from our department. Almost all cases in the literature<sup>3)</sup> resulted in secondary portal hypertension, and ligation of the hepatic artery or additional systemic portacaval shunting was used as a treatment for these patients. The results of these treatments were uncommendable because of not only palliative but also additional creation of morbid condition by systemic shunting. Direct intervention to A-P fistula should be the choice for the treatment. However, it needs wide resection of liver parenchyma because of blind situation of A-V fistula during surgery, and this operation is great burden for the patient especially with liver

dysfunction such as liver cirrhosis, causing to hepatic failure. In this report, a case of a bulky A-V fistula in the anterosuperior region of the right hepatic lobe was presented. In this case, A-V fistula in the cirrhotic liver was successfully resected by a limited partial hepatectomy under using intraoperative ultrasonographic guidance. We emphasize the usefulness of intraoperative ultrasonographic evaluation in this disease.

### Case Report

A 51-year-old woman, housewife, was admitted on 14th June, 1980 to our clinic with the chief complaints of severe hematemesis and melena. Her past history included that abnormal liver function was noted on examination in October 1980 for after-effect of exposure to the atomic bomb, and chronic hepatitis was diagnosed by SILVERMAN's needle liver biopsy on 8th October 1980. Ascites began appearing from June 1983, upon which the patient was given a diuretic. Starting in about October 1983, the patient began noticing tarry stool, and clinical laboratory tests showed mild iron deficiency anemia. On January 14th of the year, the patient had severe hematemesis and melena during the night, and was brought to our hospital.

### Clinical Record

Although the patient was in shock, but she was conscious and not jaundiced. Rapid fluid replacement restored her blood pressure to 128/78mmHg and pulse rate to 96/min. An emergency endoscopy revealed a round, bulging blood clot on the posterior wall of the stomach immediately below the cardiac orifice. It seemed to be the focus of the hemorrhage. Esophageal varix was not detected. There were no ulcerative changes in the both of the stomach and esophagus.

The abdomen was swollen. The liver and spleen were palpable below the costal margin in three and four fingerbreadths, respectively. Ascites and edema of the lower extremities were also present.

### Laboratory Findings on Admission

Blood examination gave RBC  $1.87 \times 10^4$ , hemoglobin 4.4g/dl, hematocrit 17.3%, WBC 5,200, and platelets  $10.8 \times 10^3$ ; serum biochemistry showed total bilirubin of 0.7mg%, GOT 38 SFE, GPT 26 SFE, alkali phosphatase 8.4 KA, cholinesterase 0.39 $\mu$ pH, BUN 123 mg/dl, and T.P. 5.6 g/dl. Table 1 represents the results of preoperative laboratory data after ten bottles of blood transfusion.

### Radiological and Endoscopic Studies

Upper digestive tract series showed rosette-like varices surrounding the cardiac orifice (Fig. 1). Repeated endoscopy with GTF-P3 disclosed a low bulge at a spot on the anterior wall of the esophagus about 30cm from the incisors, but the color was the same as the surrounding mucosa. Beyond the E-C junction, a typical gastric varices were seen in the lesser curvature. However, this time it was not clear where the hemorrhagic focus was. The varix was blue, without R-C signs.

On the selective celiac angiography, the liver was almost in normal size. The right anterosuperior branch of the hepatic artery was slightly enlarged, and communicated with

a blanch of the portal vein which showed a cystically enlarged termination. Hepatic artery-portal vein fistula was diagnosed (Fig. 2). In venous phase, splenomegaly was apparent but without demonstrable splenic vein. A huge varicous-like venous dilatation

**Table 1.** Preoperative laboratory data after ten bottles of blood transfusion

【Biochemical Labo. data.】 14/Fcb			
Blood (CBC)		Blood chemistry	
RBC	$331 \times 10^4/\text{mm}^3$	GOT	50 IU
Hb	10.9g/dl	GPT	25 IU
Hct	36.3%	LDH	794 IU
WBC	$2300/\text{mm}^3$	A $\beta$ -p	7.9 KA
Platelet	$6.8 \times 10^4/\text{mm}^3$	$\gamma$ -GTP	22 n
Bleed. T	2 min	TTT	11.2
Coag. T	8 min	ZST	17.0
Proth. T	12.1 sec(12.2)	T-Bil	1.9 mg/dl
ESR	28/62	Ch-E	0.46 $\Delta$ PH
HB-Ag	(-)	T-choI	94 mg/dl
HB-Ab	(-)	T.P	7.2 g/dl
CEA	2.0 ng/ml	$\gamma$ -glob	34.6%
AFP	3.2 ng/ml	BUN	12.8
ICG-Rmax	0.89	Creatinine	0.90
KICG(0.5mg)	0.11	TPHA	(-)



**Fig. 1.** Preoperative upper G.I. Series.  
Rosetlike varices surrounding the cardiac orifice (arrow mark)

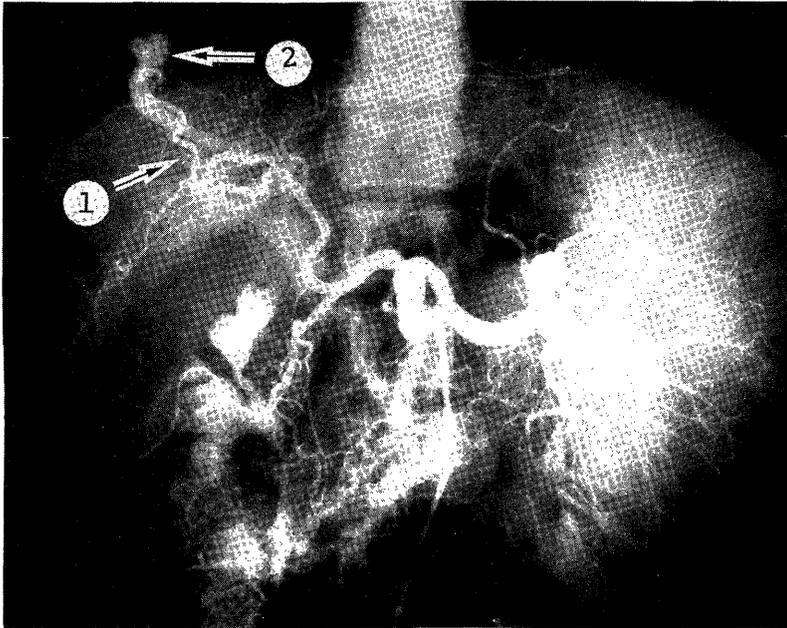


Fig. 2. Preoperative celiac angiography in the early arterial phase showing the slightly enlarged anterior superior segmental branch of the right hepatic artery (arrow mark 1) and ending in the rapid filling of a branch of the portal vein (arrow mark 2).

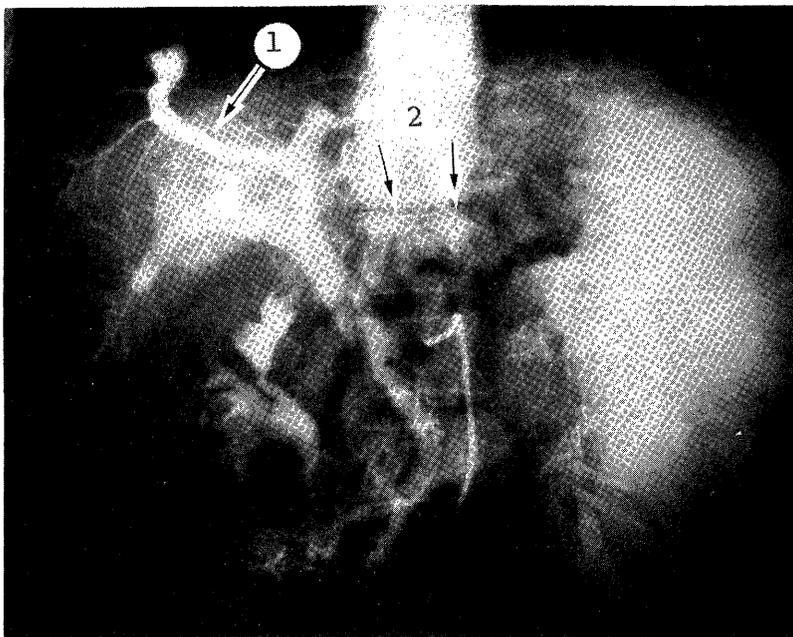


Fig. 3. Venous phase of selective celiac angiography. Reversed blood flow in the portal vein was seen apparently (arrow mark 1). Tortorous varicous venous dilatation extending along the transverse line in the upper abdomen coincidental with the course of the short gastric, the right and left gastric veins (arrow mark 2) was present.

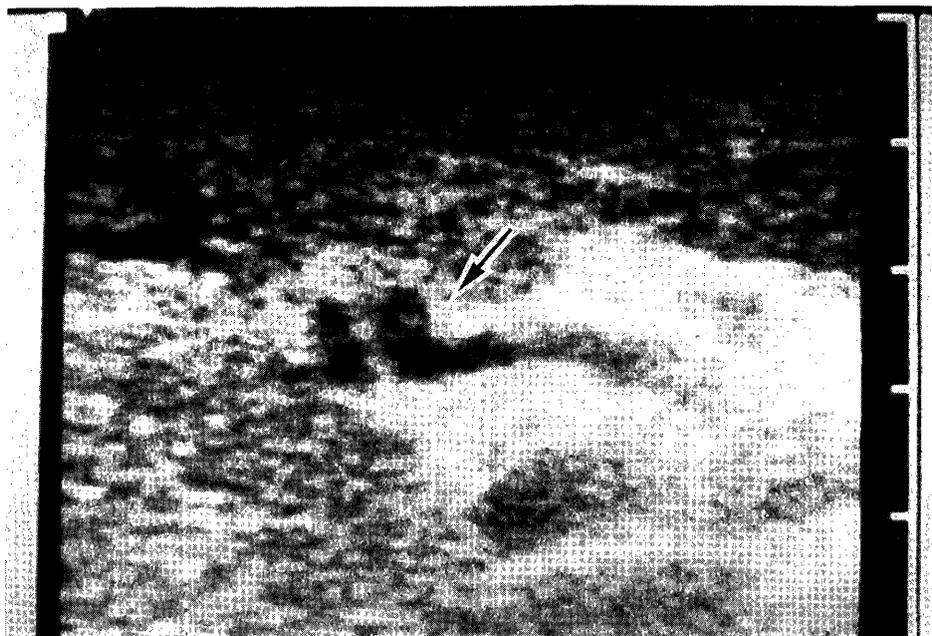


Fig. 4. Intraoperative echogram. Arrow mark showed the site of A-V fistula where the portal vein was enlarged cystically.

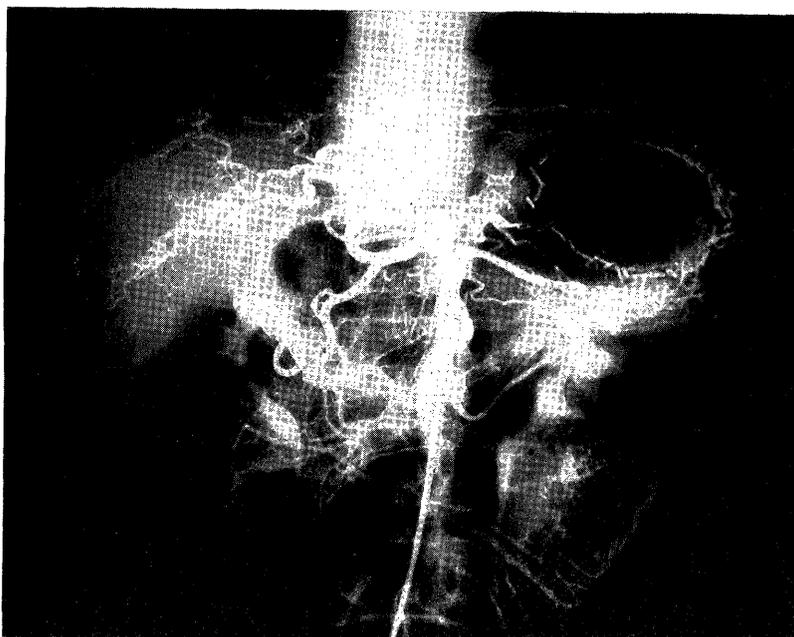


Fig. 5. Postoperative selective celiac angiography.  
A-V fistula was completely disappeared after operation.

extending from the hilus lienis to the lesser curvature of the stomach. These findings suggested a possibility of presence of shunt formation between portal system and somewhere of retroperitoneal or systemic circulation such as renal vein but not enough to relieve the increased portal blood flow through the A-V fistula.

Laparotomy was performed on February 22, 1984. Development of Hepatofugal collateral circulation through the abdominal wall, and marked dilatation of veins meandering from hepatoduodenal ligament to the hilum lienis via the lesser curvature of the stomach were evident. The liver appeared typically cirrhotic and ascites was present.

By separating the right hepatic coronary ligament, the right hepatic lobe was adequately flipped over, then operative ultrasonography was applied on liver surface to confirm the site of A-P fistula. At the first, a dilated portal branch extending from the transversal branch of the portal vein was identified, then followed this branch to its peripheral portion, and confirmed the site of A-P fistula 3 cm beneath the surface of the hepatic dome where the portal vein was enlarged cystically (Fig. 5). Under echo guidance, contrasting medium was injected into the branch of the portal vein close to the A-P fistula using a 21-gauge needle and confirmed reversed blood flow in the portal vein. Using this needle as a guide, the dilated branch of the portal vein was isolated and ligated, then the A-P fistula was successfully resected by a partial hepatectomy. The postoperative course was favorable except mild retention of ascites. But it was readily controlled by administration of diuretics. From the fifth postoperative day, the patient was allowed to eat and she was discharged from the hospital on the 14th postoperative day.

Postoperative G. I. series on the 7th postoperative day showed complete disappearance of the gastric varices which were also confirmed by gastric endoscopy.

Angiography performed on the 30th days after surgery also confirmed complete excision of the A-P shunt that had been in the right anterosuperior region of the liver (Fig. 6). In venous phase, any collateral blood flow was not present from the line of severance in the upper gastric region toward the cardiac orifice. However, a small shunt formation between splenic vein and the left renal vein became apparent at the site of confluence of splenic vein and gastric veins which had not been obvious on the preoperative selective celiac angiography.

Macroscopic examination of the specimens of resected liver showed cirrhotic liver. Microscopic examination showed that individual liver cells had relatively large irregular shapes vesicles containing coarse acidophilic substances. The matrix at some parts had widened and proliferation of bile tubules and inflammatory infiltration, mostly of lymphocytes, was evident.

## DISCUSSION

Hepatic arteriportal fistula is a disease that typically increase the portal blood flow<sup>110)</sup>. However, increased portal blood flow is not always cause portal hypertension. SIDERYS *et al.*<sup>9)</sup> constructed an aortic anastomosis to the portal vein in dogs with normal

liver and he concluded that more than 4 times increase of portal blood flow was essential for development of portal hypertension. SATO<sup>8)</sup> made a bypass between femoral and splenic arteries, and forced to increase the portal blood flow by using a roller pump. In his study, the portal pressure did not increase when the liver was functioning normally, but increased to a maximum of 445 mmH<sub>2</sub>O when the branches of the portal veins were blocked with silica grains. These two experimental studies clearly suggested that some degree of intrahepatic portal vein block, which will not enough to cause portal hypertension by itself, is basically required for the development of portal hypertension even in the circumstance of extremely increased portal blood flow which may occur in A-P shunt in man.

Presence of cirrhosis might be sure a basic factor for development of portal hypertension in this patient. However, the facts of development of ascites and severe hematemesis after the liver biopsy strongly suggested that rapid increase in portal pressure should be caused by reversed blood flow into the portal vein due to the formation through the A-P fistula which was developed by liver biopsy.

**Table 2.** Reported cases in literatures.

Causes of A-P Fistula (1892, Sacks~1983)	
Congenital	5
Ruptured aneurysm	4
Iatrogenic	9 (2)
Post-traumatic	27 (5)
Others	2 (1)
	47 (8)

( ) .....reviewed in Japanese literatures.

in four, arose after a trauma in 27, were of other origin in two, followed by biopsy with a SILVERMAN's needle in nine (Table 2).

PREGER<sup>9)</sup> in 1967 showed by angiography that a case of a development of a intrahepatic communication between hepatic arterial and portal veins after liver biopsy for diagnostic purposes. In Japan, the first of such cases was reported in 1973 by OKUDA<sup>2)</sup> who described the formation of an A-V fistula in the anteroinferior region of the right hepatic lobe in a 35-year-old patient with cirrhosis. The fistula was found in the selective hepatic arteriography subsequent to a hepatic biopsy using SILVERMAN's needle. Another case was reported by IMOTO *et al.*<sup>4)</sup> in 1977. Both of these patients were asymptomatic and mere observation of the clinical courses was sufficient. In 1975 HARADA<sup>3)</sup> reported a 41-year-old male patient with cirrhosis who had a A-P fistula after needle liver biopsy. In his case, severe hematemesis due to rupture of esophageal varices had occurred and he discussed the clinical features in detail, diagnosis, and therapy of portal hypertension caused by a hepatic artery-portal vein fistula.

Applied treatments in the literature for the patients who had A-P fistula in the

Clinically, SACKS<sup>7)</sup> in 1892 reported on a patient who died from hemorrhage of the digestive tract and stated that upon autopsy it was found that there was formation of an arteriovenous shunt between the hepatic artery and portal vein that had caused portal hypertension and the subsequent rupture of an esophageal varix. Since then, we could find only 47 cases. They were congenital in five patients and rupture of aneurysm of the hepatic artery

liver parenchyma were additional systemic-portal vein shunt operation and/or ligation of the hepatic artery, but the results disclosed these two procedures were merely palliative. Recently, improvements in ultrasonic diagnostic devices have made it possible to recognize the structures of the deeper part of the liver during surgery. At the site of A-P shunts, enlarged blood vessels such as the portal vein to a fistula are easily detectable. By using ultrasonic diagnosis at the surgery we resected completely the A-P shunt with limited partial hepatectomy. A definite conclusion has not been established between A-P shunts and cirrhosis as to the cause of portal hypertension and gastric varices in this patient.

To prevent this type of hazardous complications of needle liver biopsy, recently we introduced the needle into the liver under ultrasonographic guidance and applied fibrin adhesive in multiple layers in the tissue gap created by tissue sampling. According to a report of this method by FUJITA *et al.*<sup>2)</sup>, mild bleeding or oozing of the blood was seen in six out of 112 liver biopsies in 110 patients but without any severe complication.

## CONCLUSION

A case of portal hypertension due to hepatic artery-portal vein fistula after Silverman needle biopsy of the cirrhotic liver was treated successfully by resection of A-P shunt under ultrasonographic guidance without any postoperative complication. We emphasized the A-P fistula in cirrhotic patient should be resected by a limited partial resection of the affected liver under operative ultrasonographic guidance.

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