



Case Report

Massive Esophageal Variceal Bleeding as the Initial Presentation of Peripancreatic Tuberculoma with Portal Hypertension

Chih-Lung Chiu¹, Yi-Jen Peng², Wei-Kuo Chang³, Tsai-Yuan Hsieh³,
You-Chen Chao⁴, Heng-Cheng Chu^{3*}

¹Division of Gastroenterology, Hualien Armed Forces General Hospital, Hualien, Taiwan

²Division of Pathology, Tri-Service General Hospital, Taipei, Taiwan

³Division of Gastroenterology, Department of Internal Medicine, Tri-Service General Hospital, Taipei, Taiwan

⁴Division of Gastroenterology, Department of Internal Medicine, Buddhist Tzu Chi General Hospital, Taipei Branch, Taipei, Taiwan

Article info

Article history:

Received: August 22, 2008

Revised: September 15, 2008

Accepted: November 14, 2008

Keywords:

Abdominal tuberculoma

Esophageal variceal bleeding

Portal hypertension

Pulmonary tuberculosis

Abstract

Intra-abdominal tuberculoma is a rare manifestation of extrapulmonary tuberculosis (TB). It is even rarer for this condition to present with massive esophageal variceal bleeding. There are a few reports of portal hypertension associated with tuberculosis. A 52-year-old man with a history of chronic hepatitis B and a 1-year history of pulmonary TB was seen with complaints of hematemesis and bloody stools. He was thrombocytopenic, and abdominal computed tomography (CT) revealed a peripancreatic mass with vascular encasement and splenomegaly. CT-guided biopsy identified the tumor as a tuberculoma. During treatment for TB with an antituberculous regimen, massive esophageal varices bleeding occurred and endoscopic variceal band ligation was performed. Recurrent bleeding subsequently necessitated an exploratory laparotomy. This unusual presentation of an intra-abdominal tuberculoma with vessel encasement with portal hypertension and splenomegaly required CT-guided biopsy for a definitive diagnosis. Surgical intervention may be required for a patient who has esophageal varices resulting from portal hypertension after standard therapy for TB. Periportal tuberculoma should be included in the differential diagnosis of pancreatic head tumor with periportal vessel encasement and portal hypertension, especially for patients without evidence of cirrhosis or with a history of pulmonary TB. (*Tzu Chi Med J* 2009;21(2):172–177)

*Corresponding author. Division of Gastroenterology, Department of Internal Medicine, Tri-Service General Hospital, 325, Section 2, Cheng-Kung Road, Taipei, Taiwan.
E-mail address: chu5583@ms55.hinet.net

1. Introduction

Peripancreatic tuberculosis is an unusual complication of tuberculosis (TB) that rarely manifests in a macronodular form. Because intra-abdominal TB presents with nonspecific signs and symptoms, and is often observed without overt gastrointestinal tract or pulmonary involvement, it presents a diagnostic challenge (1–3). A definitive diagnosis is typically made after demonstrating acid-fast bacilli in a biopsy or surgical specimen, which is often performed to rule out malignancy.

The known migratory mechanisms accounting for intra-abdominal tuberculosis are: (1) hematogenous dissemination to the abdominal viscera via the lymphatic system; (2) infectious secretions transported from the respiratory tract to the gastrointestinal tract; and (3) direct invasion from a pulmonary or extrapulmonary site (4). The symptoms of peripancreatic TB are nonspecific, and include splenomegaly, ascites, cough, diarrhea, weight loss, chills, lymphadenopathy, and fever of unknown origin (4–7).

We report a 52-year-old patient with peripancreatic TB associated with encasement of the splenic vein, portal vein, and superior mesenteric artery/vein, which was complicated by portal hypertension and esophageal variceal bleeding.

2. Case report

A 52-year-old man came to our emergency department with massive hematemesis and bloody stools that began while he was mountain climbing. He had been diagnosed with pulmonary TB 1 year previously, which was confirmed by a Ziehl-Neelsen stain. He had received antituberculous treatment with isoniazid, rifampin, pyrazinamide, and ethambutol in the respiratory outpatient department. He also had a history of chronic hepatitis B, and had been followed-up in the gastrointestinal outpatient department.

On physical examination, the man was drowsy, with pale conjunctivae and dry skin. His blood pressure was 78/47 mmHg, his heart rate was 106/minute, his respiratory rate was 20/minute, and his body temperature was 36.6°C. No tenderness or superficial vein engorgement was found on abdominal examination. The liver span was 12 cm over the right midclavicular line, and the spleen span was 5 cm below the left midclavicular line, which indicated splenomegaly.

The results of laboratory studies showed the following (Table 1): white blood cell count (WBC), 3000/mL (normal range, 3500–9100/mL); hemoglobin, 7.9 g/dL (normal range, 13.5–18 g/dL); platelet count, 30,000/ μ L (normal range, 157,000–377,000/ μ L); aspartate aminotransferase (AST), 18 IU/L (normal range, 11–39 IU/L); alanine aminotransferase (ALT), 25 IU/L (normal range, 4–38 IU/L); total bilirubin, 2.2 mg/dL (normal range, 0.2–1.0 mg/dL); ammonia, 79 μ g/dL (normal, <70 μ g/dL); creatinine, 1.2 mg/dL (normal range, 0.6–1.5 mg/dL); blood urea nitrogen (BUN), 21 mg/dL (normal range, 9–23 mg/dL); prothrombin time, 14 seconds (control, 12–15 seconds); α -fetoprotein (AFP), <3.0 ng/mL (normal, <20 ng/mL); carcinoembryonic antigen (CEA), <1.0 ng/mL (normal, <5 ng/mL); cancer antigen 125 (CA-125), 15.11 U/mL (normal, <35 U/mL); and CA19-9, 17.81 U/mL (normal, <35 U/mL). His hepatitis B surface antigen (HBs-Ag) was positive and markers for hepatitis e antigen (HBe-Ag), anti-hepatitis C (HCV), and anti-human immunodeficiency virus (anti-HIV) were all negative. His HBV viral load was approximately 194,000 copies/mL.

Abdominal ultrasound revealed marked splenomegaly (15.2 cm) and no ascites. Esophagogastroduodenoscopy revealed tortuous (F2) blue varices over the lower third of the esophagus, with red wale marking and a possible hematocystic spot, indicating recent bleeding; otherwise, no active lesion was identified to be responsible for the bleeding episode. Banding ligation therapy was performed after admission (Fig. 1). Thus, we initially suspected that the

Table 1 — Summary of the patient's serial laboratory data

Admission	2 mo ago	1 mo ago	D1	D3	D90
WBC	2030	2600	3000	2700	4900
Hb (g/dL)	11.5	11.7	7.9	8.6	12.9
PLT	51,000	48,000	30,000	55,000	263,000
AST/ALT	23/41	24/26	18/25	23/15	22/29
T. bil	0.8	0.5	2.2	1.5	0.8
Albumin	4.4	4.5	3.7	4.0	4.3
Ammonia			79		111
PT (sec)	13.4/11.6	12.8/11.9	14.0/11.5	13.1/11.5	12.6/11/1
INR	1.3	1.1	1.5	1.2	1.3

WBC = white blood cell count; Hb = hemoglobin; PLT = platelet count; AST = aspartate aminotransferase; ALT = alanine aminotransferase; T. bil = total bilirubin; PT = prothrombin time; INR = international normalized ratio.

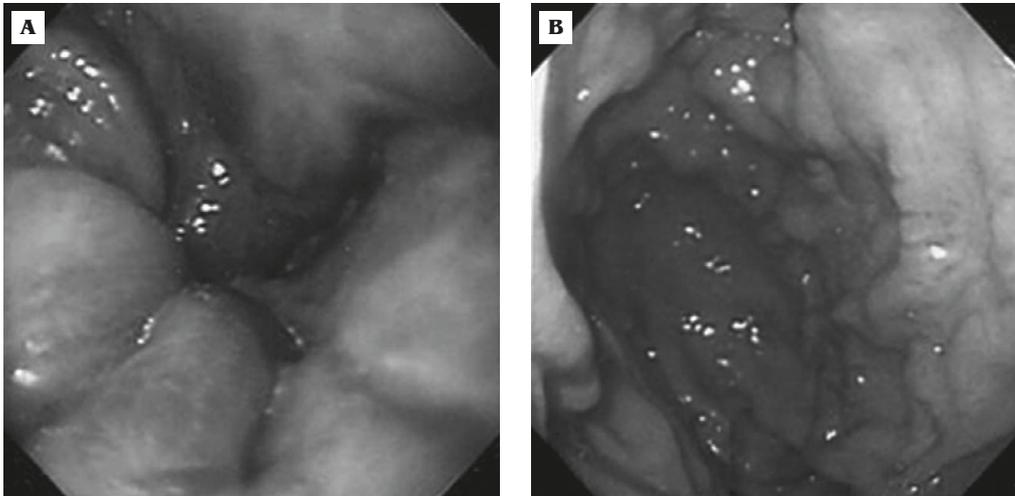


Fig. 1 — (A) Upper gastrointestinal panendoscopy shows tortuous blue varices over the lower third of the esophagus, with wale marking and a possible hematocystic spot, indicating recent bleeding. (B) Upper gastrointestinal panendoscopy also shows tortuous varices in the fundus and cardia of the stomach.

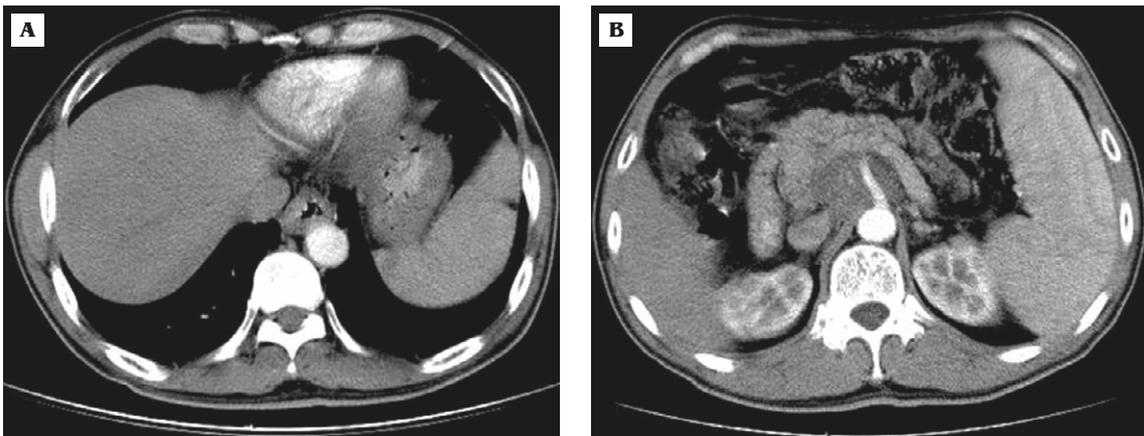


Fig. 2 — (A) Abdominal computed tomography (CT) reveals lower esophageal wall thickening and an uneven mucosal surface. (B) Abdominal CT reveals a soft tissue mass (4.2×5.6×5.0 cm) over the retro- and peripancreatic head region, causing partial encasement of the common hepatic artery and total encasement of the portal vein–superior mesenteric vein–splenic vein junction. It cannot be separated from the head of the pancreas.

patient had HBV-related liver cirrhosis, Child's classification A, with recurrent esophageal variceal bleeding. Subsequent abdominal computed tomography (CT) revealed a soft-tissue mass (4.2×5.6×5.0 cm) over the retro- and peripancreatic head region, which caused partial encasement of the common hepatic artery and total encasement of the portal vein–superior mesenteric vein (SMV)–splenic vein junction (Fig. 2B). It also showed lower esophageal wall thickening and an uneven mucosal surface compatible with esophageal varices (Fig. 2A).

Under fluoroscopic control and local anesthesia, with the patient in the prone position, CT-guided biopsy of the retropancreatic tumor was performed. The biopsy results showed chronic granulomatous

inflammation characterized by caseous-like necrosis with surrounding epithelioid histiocytes and Langhans-type giant cells (Fig. 3). Under the impression of TB, we performed an acid-fast stain but no microorganisms were found. A TB-polymerase chain reaction of the paraffin-embedded tissue yielded negative results. With this clinical history and typical pathologic findings, TB-related intra-abdominal tuberculoma should be considered first for a patient in Taiwan. Thus, a diagnosis of peripancreatic tuberculoma with vessel encasement and portal hypertension complicated by esophageal variceal bleeding was made.

After endoscopic variceal band ligation, the hematemesis resolved. However, the patient continued to have bloody stools and mild hematemesis while

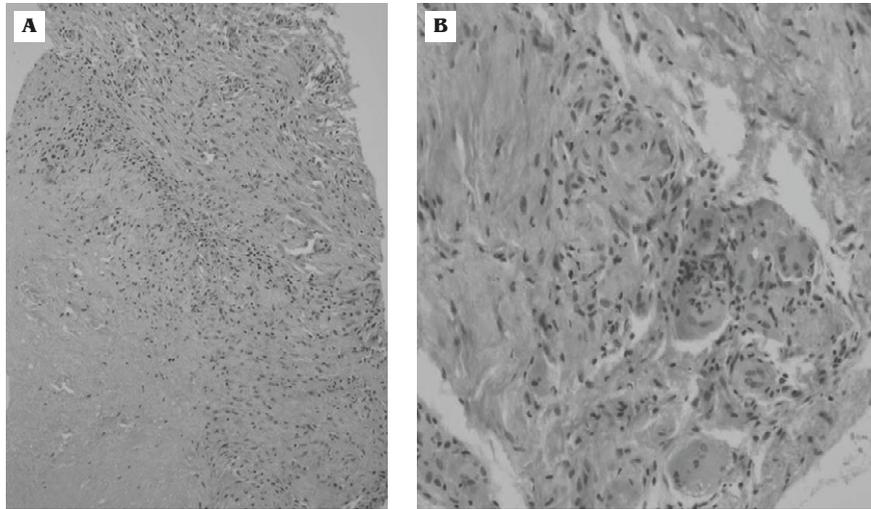


Fig. 3 — Histopathologic examination of a retroperitoneal lesion found on computed tomography-guided biopsy discloses chronic granulomatous inflammation with caseous-like necrosis and surrounding epithelioid histiocytes and Langhans-type giant cell infiltration (hematoxylin & eosin): (A) 100 \times ; (B) 200 \times .



Fig. 4 — Macroscopic operative specimen of the tuberculoma from exploratory laparotomy shows vessel sclerotic changes and encasement between the junction of the extrahepatic portal vein, superior mesenteric vein and splenic vein area.

in the hospital, suggesting that the intra-abdominal tuberculoma had not completely resolved and that vessel encasement with portal hypertension persisted.

Exploratory laparotomy was performed later because of recurrent esophageal bleeding and a poor response to medical therapy. The preoperative diagnosis was portal vein encasement with variceal bleeding, SMV sclerosis, and splenomegaly with hypersplenism. The operative findings showed vessel sclerotic changes and encasement between the junction of the extrahepatic portal vein, SMV and splenic vein area (Fig. 4). Healthy liver parenchyma without evidence of cirrhosis was found. A splenectomy, portal-systemic shunt,

splenic vein anastomosis (Warren shunt), and liver biopsy were performed.

Liver biopsy results showed chronic hepatitis, stage 1, grade 1 (Fig. 5A). There was a normal sinusoid pattern (Fig. 5B) on silver stain and a minimally enlarged portal area on Masson's trichrome stain. Microscopic examination of the splenic vein specimens demonstrated evidence of an atheroma, which was compatible with the CT-guided biopsy result. All specimens showed no evidence of malignancy. Prolonged antituberculous treatment for 6 months was instituted for this patient.

Follow-up endoscopy 3 months later revealed no esophageal varices and improvement of the gastric varices (Fig. 6). The patient currently remains in stable condition.

3. Discussion

Variceal bleeding secondary to extrahepatic portal hypertension is a different problem from variceal bleeding in patients with cirrhosis (8). In this case, the following preoperative evidence suggested the presence of a tuberculoma: (1) pre-existing pulmonary TB; (2) splenomegaly, commonly identified with intra-abdominal TB with splenic involvement (9); (3) bloody stools and hematemesis, indicating gastrointestinal tract involvement (10); and (4) portal hypertension, suggesting portal vein stenosis, upper gastrointestinal hemorrhage, increased intra-abdominal pressure (11), and liver cirrhosis (12). Of note, published data have indicated no consensus on the relationship between liver function and portal hypertension (13), further suggesting the involvement of a

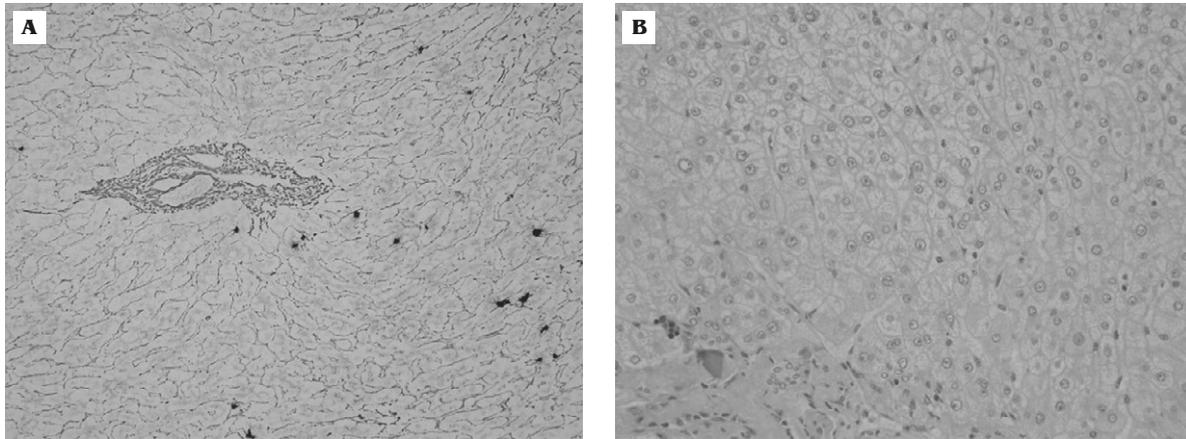


Fig. 5 — Microscopic pathology of the liver biopsy specimen shows a sinusoid pattern on silver stain: (A) 200×; (B) no cirrhosis picture is seen (400×).

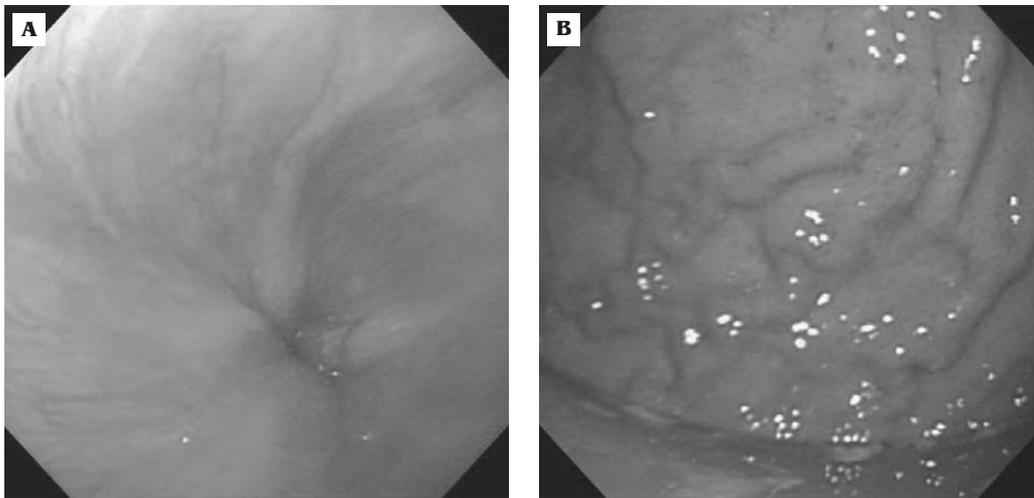


Fig. 6 — Esophagogastroscopy 3 months after surgery shows improvement of the: (A) esophageal varices; and (B) gastric varices, compared to the preoperative status.

TB-mass-encasement-induced stenosis rather than a direct HBV-related response. In this context, the esophageal varices bleeding noted during the upper gastrointestinal panendoscopy was most likely a collateral response to the increase in the portocaval pressure gradient.

Diagnosing peripancreatic/splenic TB is difficult, and differentiating peripancreatic TB from peripancreatic tumors and carcinoma is also complex (14). The possibility of cirrhosis or malignancy-induced portal hypertension can be eliminated by examining hepatocellular function with a Modified Child-Pugh score or Heinz Kalk classification.

Reports in the literature indicate that pancreatic lesions infected with mycobacteria mimic cystic neoplasms of the pancreas (15). A definitive diagnosis of abdominal TB requires demonstration of acid-fast bacilli on a smear or culture, the presence of

characteristic granulomas, and radiological features compatible with TB on barium radiographs, ultrasound, or CT scans of the abdomen. CT-guided biopsy is not the only diagnostic approach for confirming intra-abdominal TB, but when the patient has a large peripancreatic mass, it is prudent to order a diagnostic biopsy to demonstrate acid-fast bacilli and to exclude malignancy. CT-guided noncoaxial fine-needle aspiration biopsy with an approach that crosses the inferior vena cava or renal vein is safe and effective for obtaining diagnostic specimens from pancreatic and peripancreatic masses, and has been found to have an overall diagnostic accuracy of 86%.

This case report illustrates a rare cause of esophageal variceal bleeding in a patient with chronic viral hepatitis and pulmonary TB. Most clues, including esophageal variceal bleeding, splenomegaly and thrombocytopenia, and positive stains for HBsAg, are thought

to point to portal hypertension-related liver cirrhosis due to HBV. Despite this, one should also consider the length of infection with viral hepatitis and the viral load. The possible effects of hepatotoxic agents such as antituberculous drugs should also be taken into account.

Normal findings for prothrombin time, albumin, liver function tests and liver biopsy can be misleading in these cases. It is important to remember that portal hypertension can be divided into three types: prehepatic, intrahepatic and posthepatic hypertension.

Lee et al reported a 27-year-old man who suffered from portal hypertension with esophageal variceal bleeding. However, the portal hypertension was caused by periportal tuberculous lymphadenitis rather than a peripancreatic tuberculoma with vessel encasement (16). Our case illustrates the need to include intra-abdominal TB in the differential diagnosis of patients with a history of pulmonary TB who present with an abdominal mass, vascular encasement, and splenomegaly.

4. Conclusion

We believe that periportal tuberculous lymphadenitis should be included in the differential diagnosis of extrahepatic portal hypertension, especially for patients without evidence of cirrhosis or with a history of pulmonary TB. It has a poor response to a standard antituberculous regimen and early surgical intervention may prevent acute complications. The condition is very rare, but is curable with proper management.

References

1. Levine C. Primary macronodular hepatic tuberculosis: US and CT appearances. *Gastrointest Radiol* 1990;15:307-9.
2. Leder RA, Low VH. Tuberculosis of the abdomen. *Radiol Clin North Am* 1995;33:691-705.
3. Xia F, Poon RT, Wang SG, Bie P, Huang XQ, Dong JH. Tuberculosis of pancreas and peripancreatic lymph nodes in immunocompetent patients: experience from China. *World J Gastroenterol* 2003;9:1361-4.
4. Woodfield JC, Windsor JA, Godfrey CC, Orr DA, Officer NM. Diagnosis and management of isolated pancreatic tuberculosis: recent experience and literature review. *ANZ J Surg* 2004;74:368-71.
5. Winternitz MC. Tuberculosis of the spleen. *Arch Intern Med* 1912;9:680-97.
6. Piersimoni C, De Sio G, Petroni S, et al. *Mycobacterium avium* and multi-drug-resistant *Mycobacterium tuberculosis* disseminated mixed infection in a patient with acquired immunodeficiency syndrome. *Clin Microbiol Newsletter* 1995;17:6-7.
7. Bora P, Gomber S, Agarwal V, Jain M. Splenic tuberculosis presenting as hypersplenism. *Ann Trop Paediatr* 2001;21:86-7.
8. Galloway JR, Henderson JM. Management of variceal bleeding in patients with extrahepatic portal vein thrombosis. *Am J Surg* 1990;160:122-7.
9. Lundstedt C, Nyman R, Brismar J, Hugosson C, Kagevi I. Imaging of tuberculosis. II. Abdominal manifestations in 112 patients. *Acta Radiol* 1996;37:489-95.
10. Marshall JB. Tuberculosis of the gastrointestinal tract and peritoneum. *Am J Gastroenterol* 1993;88:989-99.
11. Luca A, Cirera I, Garcia-Pagan JC, et al. Hemodynamic effects of acute changes in intra-abdominal pressure in patients with cirrhosis. *Gastroenterology* 1993;104:222-7.
12. Toyonaga A, Iwao T. Portal-hypertensive gastropathy. *J Gastroenterol Hepatol* 1998;13:865-77.
13. Orloff MT, Orloff MS, Orloff SL. Treatment of bleeding from portal hypertensive gastropathy by portocaval shunt. *Hepatology* 1995;21:1011-7.
14. Kapoor VK. Abdominal tuberculosis. *Postgrad Med J* 1998;74:459-67.
15. Liu Q, He Z, Bie P. Solitary pancreatic tuberculous abscess mimicking pancreatic cystadenocarcinoma: a case report. *BMC Gastroenterol* 2003;3:1.
16. Lee CW, Lee YS, Cho GY, Kim JY, Min YI. A case of extrahepatic portal hypertension caused by periportal tuberculous lymphadenitis. *J Korean Med Sci* 1994;9:264-7.