

Primary Tubercular Liver Abscess Complicated by Tubercular Meningitis in Portal Cavernoma Cholangiopathy

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ABSTRACT

While hepatic tuberculosis is rare, primary tubercular liver abscess (TLA) is a rarer condition even in endemic countries such as India. Liver abscess in portal cavernoma cholangiopathy (PCC) is predominantly pyogenic. A 14-year-old girl was found to have PCC with multiple liver abscesses. Persistent fever and development of neurological symptoms prompted further evaluation, and she was found to have primary TLA complicated by tubercular meningitis. We report a rare case of primary TLA complicated by tubercular meningitis in asymptomatic PCC.

INTRODUCTION

Hepatic tuberculosis (TB) is a less common form of extrapulmonary TB.¹ Primary tubercular liver abscess (TLA) is an extremely rare condition, even in countries such as India, where TB is highly prevalent.^{2,3} Liver abscesses in portal cavernoma cholangiopathy (PCC) are predominantly pyogenic due to associated cholangitis. To our knowledge, this is the first report of primary TLA in a patient with PCC.

CASE REPORT

A 14-year-old girl presented with a history of fever, abdominal pain, and anorexia for 1 month. Fever was intermittent and associated with chills. Abdominal pain was continuous, described as a dull ache in the epigastric and right hypochondriac regions. On physical examination, she was febrile (temperature 101°F) and pale with unremarkable respiratory and cardiovascular examination. Abdominal examination showed epigastric tenderness. Liver was palpable 4 cm below right midclavicular line, tender, firm in consistency. Spleen was palpable 2 cm below left costal margin. Routine blood investigations showed hemoglobin 8.8 g/dL, white blood cells 8,800/mm³, and platelet count 326 × 10³/μL. Liver function tests were normal except for elevated alkaline phosphatase of 750 IU/L. Renal function tests, urine examination, tests for malaria, dengue, and chest x-ray were normal. Blood culture and HIV tests were negative.

She underwent ultrasonography (US) of abdomen with portosplenic Doppler, which showed a 4.5 x 3.5 cm ill-defined hypoechoic collection in segment V and VII of liver with another 2 x 3 cm collection in segments IV. Portal vein measured 14 mm with echogenic thrombus into right and left branches and segmental branches. Multiple tortuous venous collaterals were noted, showing monophasic flow and loss of phasicity replacing the portal vein, suggestive of portal cavernoma. Common bile duct (CBD) measured 8 mm with pericholecystic collaterals. Esophagogastroduodenoscopy was normal. Contrast-enhanced computed tomography of the abdomen confirmed portal cavernoma with liver abscesses as described and dilated tortuous CBD surrounded by multiple collaterals (Figure 1). Provisional diagnosis of extrahepatic portal obstruction with PCC with cholangitic liver abscess was

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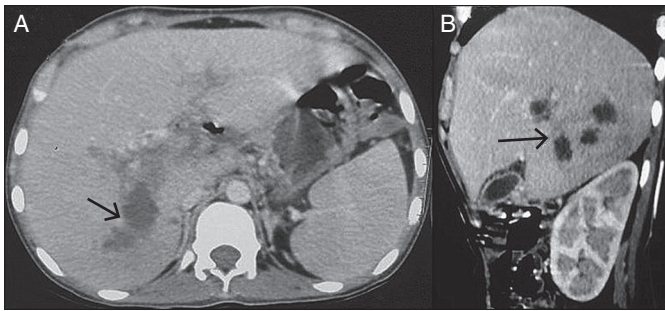


Figure 1. Contrast-enhanced computed tomography of abdomen in venous phase showed multiple abscesses in segment V and VII of liver (arrows) and multiple tortuous portal collaterals.

considered because all of the consensus criteria to define PCC were fulfilled. Negative thrombophilia profile ruled out inherited thrombophilic conditions. Gram stain of pus aspirated from the abscess was negative, and initial cultures showed no growth. For further evaluation of PCC, we performed endoscopic US, which showed prominent and tortuous CBD, with an average measurement of 8 mm and with surrounding multiple tortuous venous collaterals (Figure 2). No stone or sludge was seen in bile duct. Patient was started on antibiotics and supportive treatment. Endoscopic retrograde cholangiopancreatography (ERCP) and biliary stenting were performed considering symptomatic PCC (Figure 3). Bile culture showed no growth. She became afebrile and was discharged on oral antibiotics.

Two weeks later, she was again admitted with similar febrile episodes. Repeat US of the abdomen showed no improvement in abscess size. She was again put on broad-spectrum antibiotics. Considering nonresponse to treatment, other less

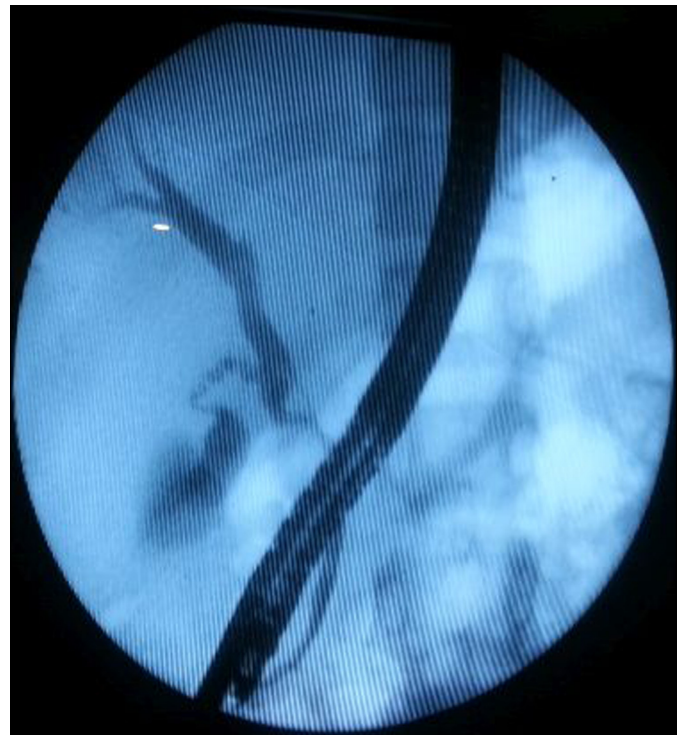


Figure 3. Cholangiogram taken during ERCP shows dilated and tortuous common bile duct with indentations and irregularities along its wall, consistent with portal cavernoma cholangiopathy.

common causes were also considered. The abscess was again aspirated and sent for bacterial culture, TB polymerase chain reaction (PCR), and mycobacterium growth indicator tube (MGIT) culture. The next day, she developed headache and double vision. Neurological examination revealed neck

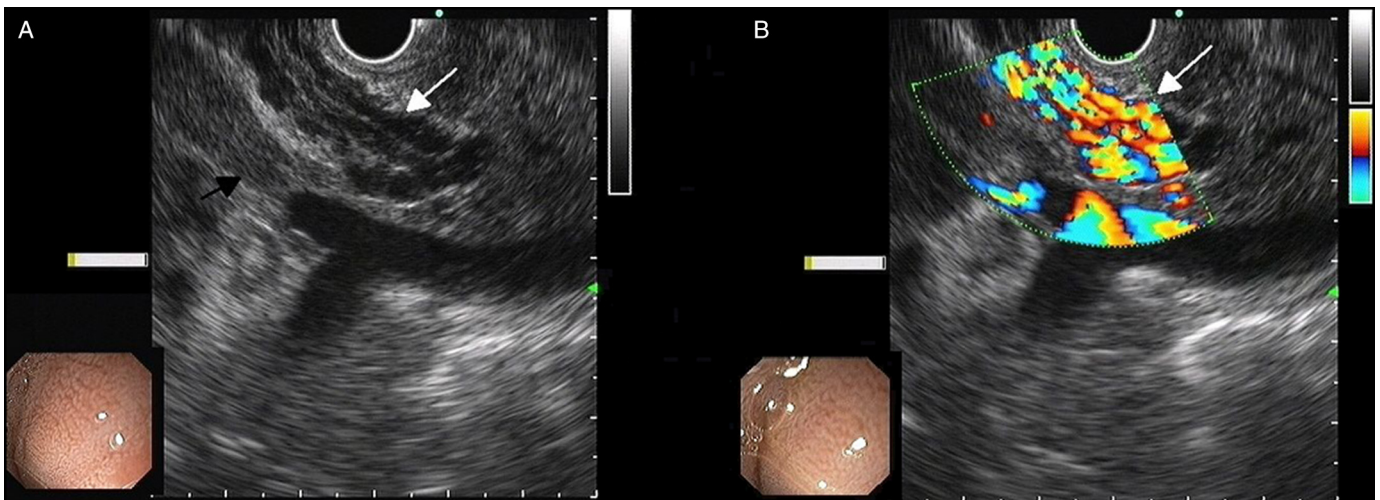


Figure 2. Endoscopic ultrasound showing thrombus distal to portal confluence with involvement of both right and left portal branches with multiple venous collaterals. (A) Dilated tortuous common bile duct with multiple pericholedochal and epicholedochal varices (arrow). (B) Doppler signals in those collaterals (arrow) confirmed our findings.

rigidity, bilateral extensor plantar response, and diplopia. Magnetic resonance imaging of brain revealed leptomeningitis of bilateral frontal sulci and few other basal areas. Cerebrospinal fluid (CSF) examination showed 300 cells/hpf (80% lymphocytes), 206 mg/dL proteins, and 35 mg/dL sugar. The TB PCR of her liver abscesses isolated mycobacterium TB, which was confirmed by positive MGIT culture at a later date. CSF TB PCR was also positive for mycobacterium TB.

She was put on injectable dexamethasone for meningitis and four antitubercular drug therapies (rifampicin, isoniazid, pyrazinamide, and ethambutol). Neurological symptoms regressed within a week and she became completely afebrile. The plastic stent was removed. She had significant improvement in appetite and gained 3 kg weight in 1 month. Abscess size reduced in follow-up US at 1 month.

DISCUSSION

Hepatic TB has varied presentations. Generally, TB spreads to the liver either from the lung via the hepatic artery or from the gastrointestinal tract via the portal vein.⁴ There are three types of hepatic TB: diffuse hepatic involvement with miliary or pulmonary TB; primary miliary TB of liver; and focal lesion in the liver, presenting as a nodule or abscess. Our patient's case fits the third type. Isolated TLA is the rarest among them.⁵ In one study, TLA was seen in only 0.34% patients with hepatic TB.⁶ TLA is commonly confused with pyogenic amoebic abscess or hepatoma. The definitive diagnosis can only be made on histological and bacteriological workup. Mycobacterium tuberculosis in pus is positive in only 0–45% of cases.⁷ Cultures can be positive in only 10% of patients.⁷ PCR is a useful diagnostic tool that was positive in 57% of patients in one study.⁸ Management of TLA consists of antitubercular therapy alone or combined with percutaneous drainage⁹ and transcatheter infusion of antitubercular drugs.¹⁰ Surgery is indicated in unsuccessful percutaneous aspiration or multiseptate abscesses.¹¹

Extrahepatic portal venous obstruction (EHPVO) is the cause of portal hypertension in 40% of adults and 80–85% of children in India.¹² The frequency of PCC in EHPVO is 81–100% in various studies.¹³ According to consensus statement guidelines, PCC is defined as abnormalities in the extrahepatic biliary system including the cystic duct and gallbladder with or without abnormalities in the 1st- and 2nd-generation biliary ducts in a patient with portal cavernoma. To diagnose PCC, the following three criteria should be fulfilled: presence of portal cavernoma, cholangiographic changes on ERCP or magnetic resonance cholangiopancreatography consistent with typical changes described for PCC (irregularity, undulation, scalloping, smooth extrinsic nodular, spiral, or stenotic impressions, and filling defects), and absence of other causes of these changes like primary sclerosing cholangitis, bile duct

injury, or cholangiocarcinoma.¹⁴ Our case fulfilled all three criteria. Most patients are asymptomatic and are incidentally found to have biliary abnormalities on cholangiography.¹⁴ Only 5–38% of patients present with symptoms.¹⁵ PCC can be diagnosed by US with color Doppler as the initial imaging modality. Asymptomatic PCC does not require any treatment, while symptomatic PCC should be treated either by endoscopic or surgical modalities. Plastic stenting with repeated and timely stent exchanges is the first-line intervention for jaundice or cholangitis due to biliary strictures in PCC. Bile duct stones are also removed with ERCP.¹⁶ Endoscopic US appears to be the method of choice to investigate varices in the CBD wall prior to ERCP. It helps to plan the procedure and to manage anticipated complications such as hemobilia.¹⁷

Our patient's elevated alkaline phosphatase, presence of multiple liver abscesses, portal cavernoma, and biliary abnormalities on ERCP along with fever and abdominal pain were consistent with the diagnosis of PCC with cholangitic liver abscess. The lack of resolution of symptoms after biliary stenting and development of meningitis prompted us to think of uncommon causes. The chronic clinical course was in favor of TLA. Development of tubercular meningitis and diplopia strengthened our diagnosis. TLA was diagnosed with TB PCR and MGIT culture and tubercular meningitis with CSF TB PCR. The patient was started on an antitubercular regimen for 6 months, and symptoms resolved in 1 month.

TLA, like other abdominal infections, can cause acute portal vein thrombosis, eventually resulting in portal cavernoma and PCC. However, formation of cavernoma takes between 3 weeks and 3 months, and occurrence of PCC requires longstanding portal cavernoma in the biliary and peribiliary region, causing compression and ischemia.¹⁸ PCC was not attributed to TLA in our case because of short symptom duration (1 month) and very high prevalence of other causes of EHPVO like idiopathic, neonatal, and childhood sepsis seen in this age group in India.

DISCLOSURES

Author contributions: R. Patel wrote the manuscript, supplied images, and is the article guarantor. D. Choksi, P. Poddar, M. Ingle, and P. Sawant edited the manuscript. K. Shah edited the images.

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