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Primary Hepato-Biliary Tuberculosis Mimicking Cholangiocarcinoma: Case Report

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Abstract:
Tuberculosis (TB) is a worldwide major health problem especially in developing countries. The lungs are the most commonly affected organs, but TB may affect any organ system in the body. Biliary involvement is extremely rare (1-3). We report the case of a 58-year-old female with a history of pulmonary tuberculosis treated and declared cured 25 years earlier, and a cholecystectomy 14 years ago, who was admitted to the hospital for investigation of intermittent jaundice associated with right hypochondrium pain evolving for 1 year. Abdominal computed tomography (CT) revealed a hepatic hilum mass associated with a slight dilation of the common bile duct (CBD) and intrahepatic bile ducts (IHBD). Cholangio-MRI showed a thickening of the CBD responsible for upstream bile ducts dilatation, associated with hilar lymphadenopathies. Bilio-pancreatic echo-endoscopy (EUS) revealed a mild inflammatory thickening in the mid and proximal CBD with multiple lymphadenopathies. Histopathological examination after an EUS-guided fine needle biopsy (EUS-FNB) of a hilar adenopathy identified epithelioid and gigantocellular granuloma compatible with tuberculosis.

Keywords: *Tuberculosis, Jaundice, Biliary tract, granuloma*

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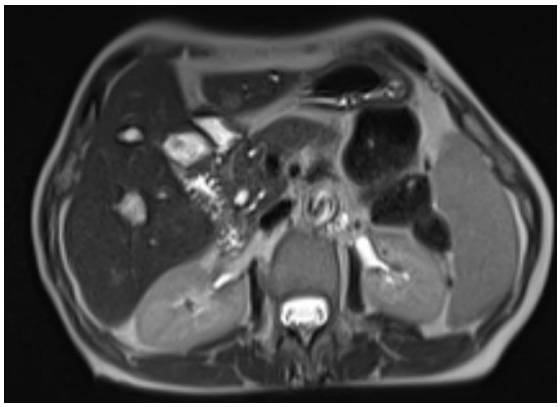
I. Introduction:

TB is a worldwide health problem with a high prevalence, approximately 95%, in developing countries (4). The lungs are the most commonly affected organs, but TB may affect any organ system in the body. The abdomen is one of the most common extrapulmonary sites of infection (5), but biliary involvement remains extremely rare. It can stem from three primary pathways: firstly, the most common way involves the dissemination of caseous material from the portal tracts into the bile ducts; secondly, it can result from secondary inflammation-related tuberculous periportal lymphadenitis; and finally, it may occur through the propagation of caseous material via the ampulla of Vater, subsequently ascending along the common bile duct (6). We aimed to report a case of primary biliary tuberculosis.

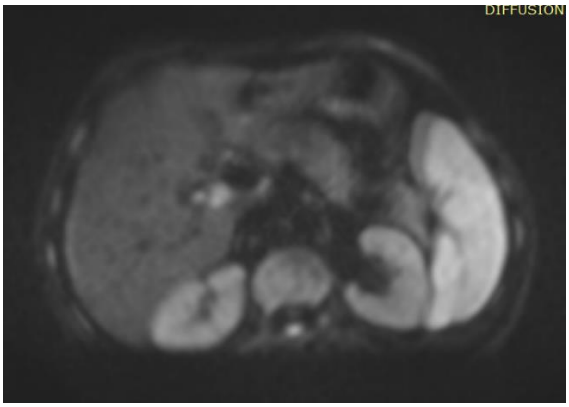
II. Case Presentation:

We report the case of a 58-year-old female patient with a medical history of treated pulmonary tuberculosis declared cured 25 years ago and a cholecystectomy performed 14 years ago, who presented for intermittent jaundice associated with right hypochondrium pain evolving over the past year. In habitual history, the patient reported taking no hepatotoxic drugs or plants, no fever, no pruritus, and no other associated symptoms apart from night sweats. Our physical examination revealed an underweight, apyretic, anicteric patient with no signs of hepatocellular insufficiency or portal hypertension, abdominal exam showed no abnormalities. Other examinations were normal. The laboratory tests, which were initially disturbed with mild cytolysis and cholestasis at 2.5 times the upper limit of normal (ULN) for PAL and GGT, normalized spontaneously. Viral serologies for B, C and HIV were negative. The complete blood count showed lymphopenia at 800 elements/ mm3. Abdominal CT revealed a hepatic hilum mass associated with a slight dilation of CBD and IHBD suggestive of cholangiocarcinoma, two hypodense hepatic lesions each measuring 1cm and enhanced after contrast agent injection compatible with hepatic angiomas, subcentimetric coeliomesenteric lymph nodes, and a mild homogeneous splenomegaly. Cholangio-MRI showed a thickening of the CBD responsible for upstream bile ducts dilatation, associated with hilar lymphadenopathies suggesting in the first instance a cholangiocarcinoma, and two hepatic lesions T2 hyperintense without diffusion restriction compatible with small hepatic angiomas. Bilio-pancreatic EUS revealed mild inflammatory thickening in the mid and proximal CBD with multiple aortocaval and hilar lymphadenopathies measuring for the largest 13x14mm. EUS-FNB was done, histopathological

examination identified epithelioid and gigantocellular granuloma compatible with tuberculosis. The tuberculin skin test (TST) was positive,the induration was 21mm of diameter. Sputum smear microscopy did not reveal any acid-fast bacilli (AFB), and culture came back negative. Xpert MTB/RIF assay on sputum samples was also negative. The chest X-ray revealed an inhomogeneous, poorly limited, non-sythematized, retractile condensation of the left upper lobe, probably sequelae. Thus, the diagnosis of isolated biliary tuberculosis was made, and patient placed under four drug regimen (isoniazid, rifampin, pyrazinamide, and ethambutol) of antibacillary treatment.



A. Axial section of a Cholangio-MRI showing dilatation of the CBD on the T2 sequence



B. Axial section of a Cholangio-MRI showing hilar adenopathy on the diffusion sequence

C. EUS images showing hilar lymphadenopathies

III. Discussion:

Hepatobiliary TB is a rare manifestation of Mycobacterium tuberculosis infection. It is more common in males with a male to female ratio of 2:1 and there is no specific age group but according to one study the majority of patients fall within the age range of 11-50 years (7).

Biliary tree contamination may be caused by 3 different processes; the spread of caseous material from the portal tracts into the bile ducts, dissemination from periportal adenitis, and ascending spread of caseous material through the ampulla of Vater (8). The disease can affect both small- and large-caliber bile ducts, possibly leading to the development of ductal strictures over time. These strictures typically manifest in a multisegmental pattern, often resulting in subsequent parenchymal atrophy of the liver segments affected.

The clinical presentation of biliary tuberculosis is slow and insidious, typically indistinguishable from malignancy. In most of the series right upper quadrant or non-specific abdominal pain appeared to be the most common symptom present in 65-87% of patients (7,9). The presence of jaundice suggests potential biliary stricture.

Liver function tests including aspartate aminotransferase (ASAT), alanine aminotransferase (ALAT), alkaline phosphatase (ALP), gamma-glutamyltranspeptidase are often near normal and therefore not diagnostic (2). A disproportionately increased serum ALP level is a consistent finding suggestive of biliary stasis. Most authors agree that cholestasis is due either to pericholangitis, or to porta hepatis nodes causing biliary compression, or possibly by direct involvement of biliary epithelium or to the rupture of a tuberculous granuloma into the bile duct (10).

Involvement of the extrahepatic biliary system is infrequent (11). When significant involvement of larger bile ducts occurs, thickening and dilation of the ductal walls emerge as the most common imaging findings (12). Additionally, biliary tract dilatation might arise due to external mechanical compression from the enlarged periportal lymph nodes. Bile duct strictures with alternating areas of dilation and stenosis detected on cholangio-MRI images may mimic primary sclerosing cholangitis (PSC) (11,13).

There have been very few reports on the use of EUS in diagnosing biliary TB. It can be helpful by providing the appearance of lymphadenopathies, showing ductal stenosis and dilatation, and especially to rule out residual lithiasis of CBD in case of cholecystectomy. The use of EUS provide also the opportunity to obtain the materials needed for cytological and microbiological evaluations using fine needle aspiration (FNA) or fine needle biopsy (FNB) (14). We came across a single study where EUS played a role in ruling out residual biliary microlithiasis and non-oncological pathology. The patient in this case initially presented with jaundice and was undergoing treatment for both human immunodeficiency virus (HIV) and TB. Notably, the patient had a prior history of cholecystectomy for gallstones. Abdominal ultrasound did not reveal any unusual findings in the liver, such as biliary dilation or filling defects. Initially, the patient was suspected to have hepatitis due to TB medications, leading to the discontinuation of treatment. However, instead of improvement, there was a progressive rise in the levels of indirect bilirubin. Subsequently, an EUS procedure was conducted, unveiling the presence of a mass near the duodenal papilla along the distal common bile duct. Further analysis through EUS Fine-Needle Aspiration (EUSFNA) confirmed the presence of lymphocytes that tested positive for TB bacilli. The patient subsequently underwent endoscopic cholangiopancreatography with sphincterotomy and the placement of a biliary drainage endoprosthesis, and TB treatment was resumed. Following nine months of treatment, no masses were detected (15)

If laboratory tests and imaging techniques have proved unsuccessful, it possible to obtain a reliable diagnosis with biliary cytologic findings from endoscopic cholangiography.

IV. Conclusion:

Biliary TB is a rare extrapulmonary manifestation of one of the commonest infections caused by Mycobacterium tuberculosis. It is an uncommon cause of biliary stricture that can mimic cholangiocarcinoma. Hence, it should be considered within the differential diagnosis in patients presenting with cholestatic jaundice and suggestive imaging findings especially in areas where tuberculosis is highly prevalent. EUS and EUS-guided FNA/FNB may prevent unnecessary invasive diagnostic methods for biliary TB. Therefore, more patients can be precisely diagnosed and receive proper treatment using a less invasive procedure.

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