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CASE REPORT

Primary non-Hodgkin's lymphoma of the common bile duct: A case report and literature review

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Summary Hepatobiliary involvement by malignant lymphoma is usually a secondary manifestation of systemic disease, whereas primary non-Hodgkin's lymphoma of the extrahepatic biliary ducts is an extremely rare entity. We describe the case of a 57-year-old man who presented with an acute onset of obstructive jaundice and severe itching. Abdominal ultrasonography and computed tomography revealed intrahepatic and common hepatic ducts dilatation. Magnetic resonance cholangiopancreatography and endoscopic retrograde cholangiopancreatography showed a mid-common bile duct stricture. The patient was presumed to have cholangiocarcinoma of the common bile duct, and an *en bloc* resection of the tumor with Roux-en-Y hepaticojejunostomy and porta-hepatis lymph nodes dissection was performed. Histopathology and immunohistochemistry revealed a large B cell non-Hodgkin's lymphoma. The patient received six cycles of combination chemotherapy using cyclophosphamide, vincristine, prednisone, and rituximab (CVP-R) protocol, and after a 5-year follow-up he is still in complete remission. We also reviewed the cases published from 1982 to 2012, highlighting the challenges in reaching a correct preoperative diagnosis and the treatment modalities used in each case.

1. Introduction

Non-Hodgkin's lymphoma (NHL) accounts for 1–2% of all cases of malignant biliary obstruction.[1] To our knowledge and after reviewing the literature since the first case was published by Nguyen[2] in 1982, only 28 cases have been reported. Herein we present another case of primary NHL of

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the common bile duct. A literature review was conducted of the challenges in arriving at the diagnosis preoperatively and the debate around the optimal treatment modalities.

2. Case report

A 57-year-old man with no previous chronic medical illnesses presented with a 2-week history of severe itching all over his body, associated with general fatigue, malaise, and nausea, without vomiting or anorexia. He also reported having a yellowish discoloration of sclera, dark (tea color) urine, and a bulky foul-smelling stool. However, he denied any history of fever or chills, abdominal pain, previous episodes of similar symptoms, recent travels, or contact with sick people. He is not on any regular medications, and he denied recent ingestion of any drug. His surgical history and family history showed no significant finding. Moreover, he has a 40 pack-year smoking history but no history of alcohol consumption or illicit drug use.

On presentation his vital signs were within normal limits. The physical examination revealed deep scleral and skin jaundice with itching marks on his skin. There was no cervical lymphadenopathy and no stigmata of chronic liver disease. An abdominal examination revealed a soft and lax abdomen with mild tenderness over the epigastric and right upper quadrant areas, and active bowel sounds but no masses or hepatosplenomegaly.

His laboratory results showed hemoglobin, white cell count, and platelets of 15 g/dL, 7 $\times$ 10^3/mm^3, and 162 $\times$ 10^3, respectively. His serum total bilirubin, direct bilirubin, alanine aminotransferase, aspartate aminotransferase, alkaline phosphatase, gamma glutamyl transpeptidase, lactate dehydrogenase, and amylase results were 18.3 mg/dL 16.1 mg/dL, 116 U/L, 66 U/L, 161 U/L, 658 U/L, 898 U/L, and 58 U/L, respectively. His blood carbohydrate antigen 19-9 level rose to more than 1200 U/mL, and carcinoembryonic antigen was within normal levels at 0.88 ng/mL. Hepatitis A, B, and C serology was negative.

Abdominal ultrasonography revealed dilated common hepatic and intrahepatic ducts. An endoscopic retrograde cholangiopancreatography (ERCP) was subsequently performed and showed a 3-cm mid-common bile duct stricture with significantly dilated intrahepatic and common hepatic biliary ducts. The rushing cytology of the common bile duct was suspicious for malignancy. A 9-cm, 10 F stent was inserted across this for drainage (Fig. 1A).

The abdominal computed tomography (CT) scan revealed a mild intrahepatic biliary tree dilatation but was otherwise unremarkable (Fig. 1B).

Magnetic resonance cholangiopancreatography showed a marked tapered stricture at the distal common bile duct with mild to moderate dilatation of biliary tree proximally, and a normal pancreatic duct (Fig. 1C).

Based on the above presentation and investigations, a presumptive diagnosis of cholangiocarcinoma was made, and extrahepatic biliary tract excision, a Roux-en-Y hepaticojejunostomy, and porta-hepatis lymph nodes excision were performed.

A histopathologic examination of the common bile duct revealed a large B cell-type NHL with tumor-free surgical resection margins (Fig. 2), and the porta-hepatis lymph nodes showed a reactive nonspecific hyperplasia. Immunohistochemical staining was positive for CD3, CD5, CD20, CD45, BCL2, and Ki67, and negative for CD15 and CD30 (Fig. 3).

The patient was referred to the oncology clinic, where a positron emission tomography showed no evidence of malignant lesions. He received six cycles of chemotherapy using the CVP-R (cyclophosphamide, vincristine, prednisone, and rituximab) protocol. One-year follow-up chest, abdomen, and pelvis CT and whole-body positron emission tomography scans revealed no evidence of malignancy. He received six maintenance courses of rituximab (anti-CD20) and remains in complete remission 5 years from the time of diagnosis with a normal whole-body CT scan.

3. Discussion

Diffuse large B cell lymphoma (DLBCL) is the most common lymphoid neoplasm and the most common histologic subtype of NHL, accounting for approximately 25% of all cases.[3] It has an overall incidence rate of 3—7 cases per 100,000 persons per year. The incidence also increases with age (median age 64 years), and the disease appears to be

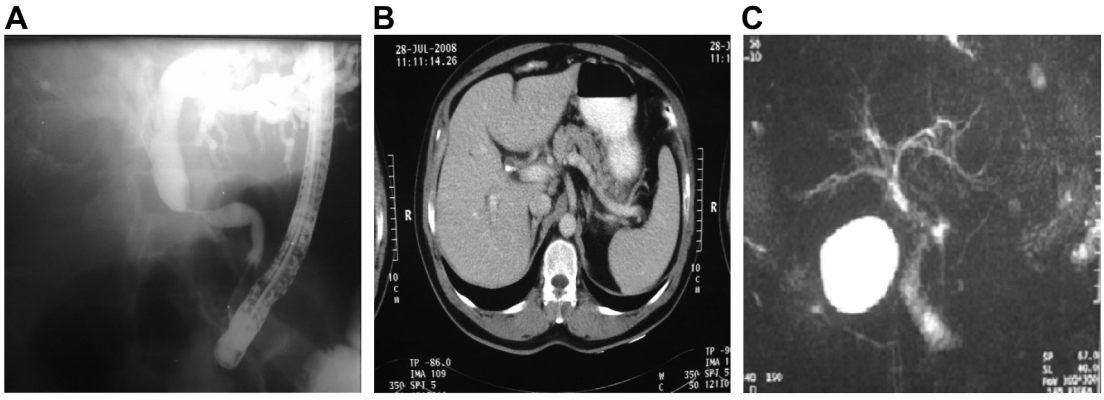

Figure 1 Imaging studies. (A) Endoscopic retrograde cholangiopancreatography showing the mid common bile duct stricture. (B) Computed tomography scan of the abdomen showing intrahepatic bile tract dilatation. (C) Magnetic resonance cholangiopancreatography showing the tapered stricture of the distal common bile duct.