

CASE REPORT

PRIMARY NON-HODGKIN'S LYMPHOMA OF THE COMMON BILE DUCT PRESENTING AS OBSTRUCTIVE JAUNDICE: A CASE REPORT

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ABSTRACT

Introduction. Primary non-Hodgkin's lymphoma of the extrahepatic bile duct presenting as obstructive jaundice is an extremely rare disease.

Case presentation. We report a case of obstructive jaundice caused by primary non-Hodgkin's lymphoma of the common bile duct (CBD), in a 67-year-old male patient. Our patient showed clinical evidence of obstructive jaundice, and tomography revealed a dilated common bile duct with the suspicion of intra ductal lithiasis. Endoscopic retrograde cholangiopancreatography (ERCP) was performed, with efficient drainage and clinical resolution of the obstructive jaundice. One month later, the patient presented the same clinical signs of obstructive jaundice, only this time the ERCP revealed common bile duct distal stenosis. The next step was to perform an echoendoscopy (EUS), that visualized a homogeneous mass at the common bile duct distal level and cholangiocarcinoma was suspected. Fine needle aspiration (FNA) of the lesion obtained tissue fragments for histological examination. The diagnosis was diffuse B-cell type non-Hodgkin's lymphoma in the marginal area, and the patient was

RÉSUMÉ

Lymphome non-Hodgkinien primaire des voies biliaires simulant un ictère par obstruction: présentation de cas

Introduction. Le lymphome primaire non-Hodgkinien des voies biliaires extra-hépatiques, qui se présente sous la forme d'un ictère par obstruction, est une maladie extrêmement rare.

Rapport du cas. Nous rapportons un cas d'ictère obstructif causé par un lymphome primitif non-Hodgkinien du canal biliaire principal chez un homme âgé de 67 ans. Notre patient a présenté des signes cliniques d'ictère obstructif et la tomographie a révélé un canal biliaire principal dilaté avec la suspicion de lithiase. Une cholangiopancréatographie rétrograde endoscopique (CPRE) a été réalisée avec un drainage efficace et une résolution clinique de la jaunisse obstructive. Un mois plus tard, le patient présentait les mêmes signes cliniques de jaunisse obstructive que cette fois-ci, la CPRE révélait une sténose distale du canal biliaire commun. L'étape suivante consistait en une écho-endoscopie (EUS) permettant de visualiser

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evaluated in the Oncology Department for chemotherapy.

Conclusions. It is very important to differentiate primary non-Hodgkin's lymphoma of the bile ducts from other causes of obstructive jaundice, as the treatment approach and prognosis are fundamentally different. Endoscopic ultrasound-guided fine-needle aspiration (EUS-FNA) is the definitive diagnosis technique, avoiding surgical interventions and the classic exploratory laparotomy.

Keywords: obstructive jaundice, non-Hodgkin's lymphoma, echoendoscopy.

INTRODUCTION

Primary non-Hodgkin's lymphoma of the extrahepatic bile duct presenting as obstructive jaundice is a rare disease¹. Only a few cases are presented in recent medical literature and mostly because non-Hodgkin's lymphoma accounts for 1-2% of all cases of malignant biliary obstruction².

CASE PRESENTATION

A 67-year-old male, hypertensive, undergoing medical treatment, is hospitalized with the suspicion of drug induced hepatitis. According to history data, the patient was treated 5 days ago for a urinary tract infection with ciprofloxacin, and 3 days after antibiotic therapy, he developed jaundice accompanied by pain in the right hypochondria, without any other symptoms (fever, pain, weight loss). His vital signs were within normal limits. The physical examination revealed scleral and skin jaundice. No superficial lymph nodes were palpable. Laboratory findings included liver cytolysis (alanine aminotransferase 808 U/L, aspartate aminotransferase 320 U/L), cholestasis (total bilirubin 8.23 mg/dL, direct bilirubin 4.51 mg/dL, alkaline phosphatase 516 U/L), hyperglycemia, slightly increased lipase (1.5x normal value). Hepatitis A, B, and C serology was negative.

The ultrasound revealed the gallbladder containing sludge, significant common bile duct dilation (15 mm in the hilum, with sudden retro-pancreatic decalibration, apparently without obstruction), non-homogeneous cephalic pancreas, the peritoneal cavity

une masse homogène du niveau distal du canal biliaire commun et un cholangiocarcinome était soupçonné. Une aspiration à l'aiguille fine (FNA) de la lésion avec des fragments de tissu et un bloc cellulaire a été obtenue pour l'examen histologique. Le diagnostic pathologique était un lymphome non-Hodgkinien de type B à cellules B diffus dans la zone marginale et le patient était évalué dans le Département d'Oncologie pour la chimiothérapie.

Conclusions. Il est très important de différencier les lymphomes primitifs non-Hodgkiniens des voies biliaires des autres causes de jaunisse obstructive, car l'approche thérapeutique et le pronostic sont fondamentalement différents. L'aspiration à l'aiguille fine sous guidage endoscopique (EUS-FNA) est dans ce cas le diagnostic définitif évitant les interventions chirurgicales et la laparotomie exploratoire classique.

Mots-clés: jaunisse obstructive, lymphome non-Hodgkinien, écho-endoscopie.

without fluid. Computed tomography was performed, confirming the ultrasound findings and alleviating the suspicion of stones in the distal biliary tract. The patient was transferred to our clinic for further investigations and specialized treatment. An endoscopic retrograde cholangiopancreatography (ERCP) was subsequently performed. The common bile duct was 12 mm, without intrahepatic bile ducts dilatation.

Efficient drainage was performed, by removing stones from the main biliary duct using the basket. Approximately one month after discharge, the patient presented with clinical evidence of obstructive jaundice. Laboratory findings included hepatic cholestasis with a total bilirubin of 5.52 mg/dL, liver cytolysis and hypocholesterolemia. Residual lithiasis was suspected. Endoscopic retrograde cholangiopancreatography revealed common bile duct distal stenosis. 10 Fr and 8.5 Fr plastic stents were placed. Due to the stenotic malignant aspect, echoendoscopy (EUS) was performed the next day. A relatively well-defined non-homogeneous mass of 22/17 mm diameter was visualized at the common bile duct distal level (Figure 1). Fine needle aspiration was performed. Peripancreatic adenopathies with a maximum size of 22 mm were also observed. The microscopic examination revealed lymphoid tissue with small lymphocytes, rounded nucleus and low cytoplasm, rare medium to large lymphocytes, rare mitosis. Necrotic tissue was absent, as well as epithelial cells. Immunohistochemistry revealed small B cells, positive tumor proliferation, positive for CD20, negative for CD5 (T marker), Cyclin D1 (B marker of mantle lymphoma) and CD10 (germinal center marker).

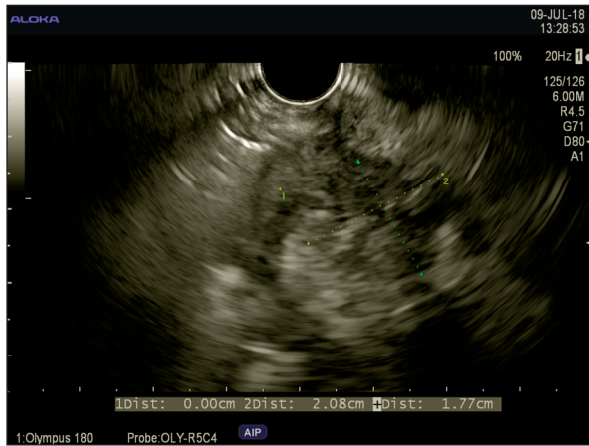


Figure 1. Non-homogeneous mass visualized by EUS at the common bile duct distal level

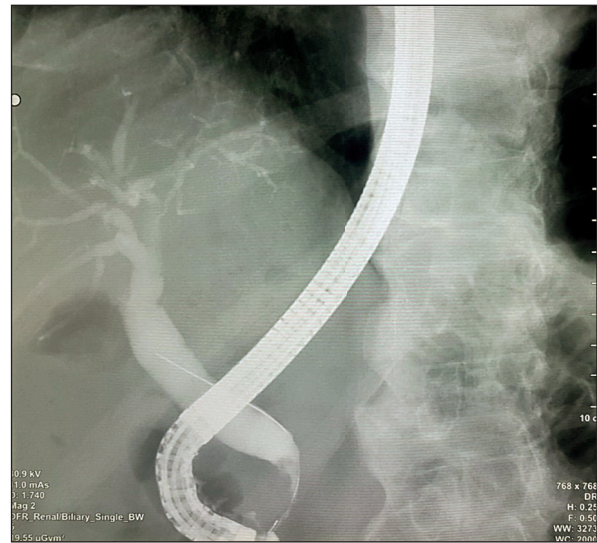


Figure 2. Common bile duct distal stenosis visualized at the endoscopic retrograde cholangiopancreatography

The Ki67 proliferation index was less than 10%. The histopathological aspect and the immunohistochemical examination are compatible with non-Hodgkin malignant B cell in the marginal area. The last endoscopic retrograde cholangiopancreatography, one month after establishing the diagnosis of certainty, revealed distal stenosis over the last 2 cm (Figure 2). Two plastic stents of 10 Fr were inserted with efficient drainage. After oncologic evaluation, combined chemotherapy including cyclophosphamide, doxorubicin, vincristine, and prednisone plus rituximab (CHOP-R) was started last month.

DISCUSSION

Diffuse large B cell lymphoma is the most common lymphoid neoplasm and the most common histologic subtype of non-Hodgkin's lymphoma, accounting for approximately 25% of all cases³. The incidence increases with age, mostly over 60 years old, and the disease appears to be slightly more predominant in men (50% of cases) than in women⁴. The gastrointestinal tract is the most common site of involvement with presenting symptoms such as abdominal pain, loss of appetite, weight loss and vomiting⁵. Obstructive jaundice is mostly secondary to compression of the extrahepatic bile ducts by periportal, perihepatic, or peripancreatic lymphadenopathy, associated tumor lysis, or direct hepatic involvement⁶. Although it remains unclear whether chemotherapy should precede the biliary drainage procedures in patients with non-Hodgkin's lymphoma presenting with jaundice, chemotherapy alone usually alleviates the obstructive jaundice without biliary drainage⁷. Dudgeon et al described five patients with non-Hodgkin's lymphoma causing obstructive jaundice, who were treated with combined chemotherapy

without prior surgical or endoscopic biliary decompression, or radiation therapy⁸. Fine needle aspiration (FNA) with endoscopic ultrasound (EUS) guidance is often required to direct therapy in the case of an uncertain diagnosis⁹. FNA with EUS guidance permits both morphologic and cytologic analysis of lesions at various locations such as within or adjacent to the gastrointestinal tract, and intra-abdominal and retroperitoneal masses¹⁰⁻¹⁵. EUS-FNA can have a role in diagnosing other lesions that may mimic cholangiocarcinoma and present either as a mass or with obstructive jaundice¹⁶⁻¹⁹. All reported cases indicate that common bile duct non-Hodgkin's lymphoma is a rapidly progressive disease and an accurate histopathologic diagnosis, fast treatment of other complications, in our case the obstructive jaundice, evaluation for surgical resection, if feasible, combined with chemotherapy with or without radiotherapy may be the approach to offer a chance for cure²⁰.

CONCLUSIONS

It is very important to differentiate the primary non-Hodgkin's lymphoma of the bile ducts from other causes of obstructive jaundice, as the treatment approach and prognosis are fundamentally different. In our case, only after the second ERCP revealed a distal stenosis of the common bile duct, EUS was performed with the suspicion of cholangiocarcinoma. The tissue biopsy by EUS-FNA, with characteristic histopathologic and immunohistochemical findings, was the gold standard for definitive diagnosis. In conclusion, although primary non-Hodgkin's lymphoma of the biliary ducts is an extremely rare disease, it

should be considered in the differential diagnosis of malignant obstructive jaundice.

Compliance with Ethics Requirements:

„The authors declare no conflict of interest regarding this article“

„The authors declare that all the procedures and experiments of this study respect the ethical standards in the Helsinki Declaration of 1975, as revised in 2008(5), as well as the national law. Informed consent was obtained from the patient included in the study“

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