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CASE REPORT

Cutaneous Large B Cell Lymphoma Involving the Duodenum and the Bile Duct: a Case Report

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Introduction

Primary cutaneous B cell lymphoma (PCBCL), leg type, is a rare and aggressive neoplasm [1, 2]. Primary or secondary bile duct obstruction is considered a poor prognostic sign for non-Hodgkin's lymphoma [3]. We present a case of a 63-year-old man, with a BCBCL, leg type, on his back, disseminated to the duodenum and the common bile duct.

Case Report

A 63-year-old male patient presented a huge indolent tumor (60 cm in diameter), with peripheral annular erythematous and pigmented reinforcement on the back next to the left shoulder, evolving for 1 year and ignored by the patient (Fig. 1). A skin biopsy was performed 3 months before admission and histologic examination showed dense diffuse lymphocytic infiltrate of centroblasts and immunoblasts. An immunohistochemical analysis showed positivity of CD20 and Bcl-2 and negativity of the other markers: MUM-1, and Bcl-6, CD30, and Epstein-Barr virus stains including Epstein-Barr encoded RNA. A primary cutaneous B cell lymphoma (PCBCL) leg type was initially established. Chemotherapy was indicated but the patient did not consult and he presented with a 2-week history of progressive obstructive jaundice and abdominal pain without fever. Physical examination showed jaundice and a palpable gallbladder but no hepatosplenomegaly or peripheral lymph nodes. His right upper abdomen was tender to palpation. Laboratory findings confirmed cholestasis with elevated levels of direct bilirubin, alkaline phosphatase, and gamma-glutamyl-transpeptidase, associated with a moderate cytolysis. Prothrombin time was normal. The cancer antigen 19-9 level was mildly elevated. Lactate dehydrogenase level was normal. Abdominal ultrasound showed dilation of the intrahepatic and common bile ducts with an enlarged gallbladder without evidence of an obstacle on the bile ducts. Abdominal CT scan and magnetic resonance cholangiopancreatography (MRCP) revealed dilatation of the proximal common bile duct (CBD) and both intrahepatic bile ducts, a narrowing of the distal common bile duct with no masses in the liver and no lymphadenopathy (Fig. 2). Pancreatic duct was also dilated. Lateral duodenoscopy showed an ulcer of the second portion of the duodenum away from the papilla; biopsies were performed. Histological examination with immunohistochemistry revealed a large B cell-type malignant lymphoma.

Endoscopic retrograde cholangiopancreatography (ERCP) and cholangiography confirmed the stricture and showed a narrowed segment in the inferior part of the CBD, with proximal dilatation (Fig. 3). After endoscopic papillotomy, cytological and biopsy tests were performed and a plastic stent was placed for drainage. Histological examination with immunochemistry showed a large B cell-type lymphoma (Fig. 4). A

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Fig. 1 Horseshoe-shaped and indurated plaque on the back

neck and chest CT scan revealed no abnormalities. Cholestatic jaundice disappeared with normalization of liver function tests. We concluded to a PCBCL, leg type, disseminated to the duodenum and to the bile duct. The patient was treated by chemotherapy (rituximab, cyclophosphamide, vincristine, doxorubicin, and prednisolone: R-CHOP). He died after 9 months of follow-up.

Discussion

PCBCL, leg type, which frequently develops on the legs of elderly patients, exhibits an unfavorable prognosis. It displays a characteristic phenotype with strong expression of Bcl-2 and MUM1, but variable expression of Bcl-6 and absence of CD10 [4]. There are different causes of jaundice in lymphoma: hepatitis, hemolysis, liver involvement, syndrome of

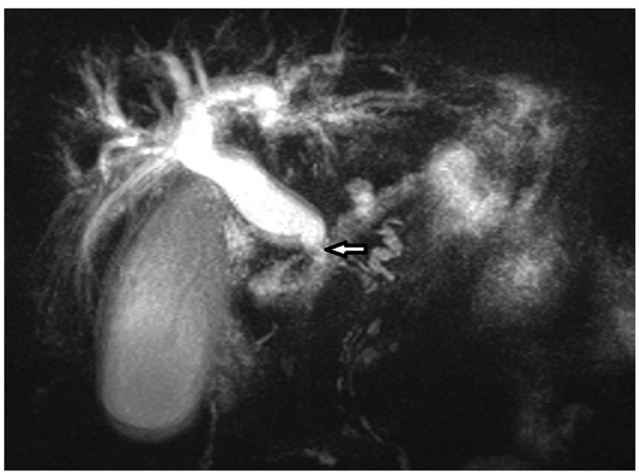

Fig. 2 Coronal magnetic resonance cholangiopancreatography (MRCP) image showing dilated proximal common bile duct and intrahepatic bile ducts with narrowing of the distal common bile duct (arrow)

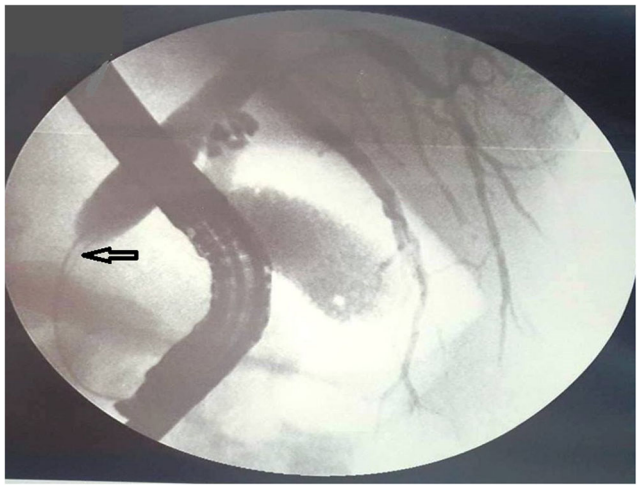

Fig. 3 Cholangiography showing stricture of the inferior part of the common bile duct (arrow)

cholestasis without liver or bile duct involvement [3, 5]. Extrahepatic bile duct obstruction by lymphoma is rare [6]. It is frequently due to compression of the extrahepatic bile ducts by lymph nodes but it may result from an infiltration of the common bile duct by the lymphoma [3, 5]. Radiologic resemblance of extrahepatic biliary lymphoma to cholangiocarcinoma in the imaging findings of CT scan and MRI was noted in the studies [7]. ERCP is more sensitive; it allows cytology brushing which may confirm the diagnosis. It also allows stent placement [6]. Moreover, the patient presented a duodenal infiltration by lymphoma. Aggressive systemic combination chemotherapy with or without involved-field radiation therapy for the cutaneous location is the recommended approach [8]. PCBCL, leg type, and primary or secondary bile duct obstruction related to lymphoma are associated with a poor prognosis [2, 3].

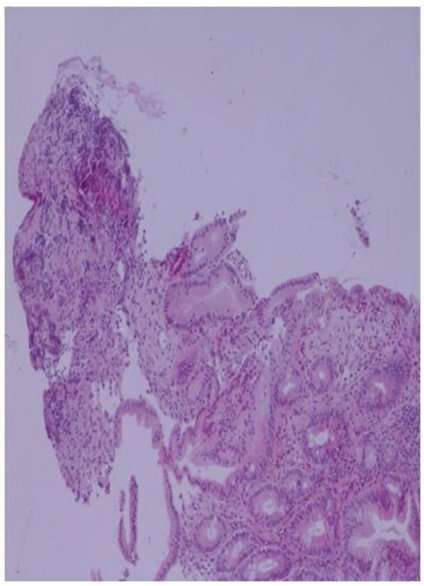

Fig. 4 Histological examination showing ampullary lymphoma

Conclusion

We have presented a case report of a rare lymphoma which is the PCBCL, leg type, with also rare disseminated sites: the duodenum and the common bile duct. This is the first case reported in the literature. The diagnosis of extrahepatic bile duct involvement remains difficult and the prognosis is poor.

Compliance with Ethical Standards

Informed Consent Informed consent was not obtained because the patient died and we have no contact of his relatives (unreachable by phone and other methods).

Conflict of Interest The authors declare that they have no conflict of interest.

Abbreviations *PCBCL*, primary cutaneous B cell lymphoma; *EBV*, Epstein-Barr virus; *EBER*, Epstein-Barr encoded RNA; *ALP*, alkaline phosphatase; *GGT*, gamma-glutamyl-transpeptidase; *AST*, aspartate amino transferase; *ALT*, alanine transferase; *PT*, prothrombin time; *CA 19-9*, Cancer antigen 19-9; *HIV*, human immunodeficiency virus; *MRCP*, magnetic resonance cholangiopancreatography; *CT scan*, computed tomography scan; *CBD*, common bile duct; *ERCP*, endoscopic retrograde cholangiopancreatography; *R-CHOP*, rituximab, cyclophosphamide, vincristine, doxorubicin, and prednisolone

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