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An Exceptional Cause of Cholestasis during Hodgkin's Lymphoma

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Abstract **Case Report**

Intrahepatic cholestasis as a paraneoplastic manifestation is an unusual phenomenon in Hodgkin's lymphoma (HL) which presents both a diagnostic problem because it must be considered after excluding all other causes of cholestasis, and a therapeutic one because the decision and timing of treatment places the clinician in a balance between the hepatotoxicity of chemotherapy and the progression of the lymphomatous disease. We report the case of a 71-years-old patient presenting with cholestatic jaundice and a significant weight loss, whose morphological and anatomopathological explorations concluded to the diagnosis of Hodgkin's lymphoma without lymphomatous infiltration of the liver, associated with idiopathic intrahepatic cholestasis without ductopenia. Low-dose chemotherapy led to clinical and biological improvement of the cholestasis.

Keywords: Hodgkin's lymphoma, Intrahepatic cholestasis, paraneoplastic manifestation.

INTRODUCTION

Intrahepatic cholestasis as a paraneoplastic manifestation is an unusual phenomenon in Hodgkin's lymphoma (HL) which presents both a diagnostic problem because it must be considered after excluding all other causes of cholestasis, and a therapeutic one because the decision and timing of treatment places the clinician in a balance between the hepatotoxicity of chemotherapy and the progression of the lymphomatous disease.

CASE REPORT

A 71-years-old man, presented with jaundice, pruritus and significant weight loss. He had no known medical history and did not take any regular medication. He had no history of smoking, alcohol consumption, drug use or any hepatotoxic substances. There was no noteworthy family history.

On physical examination, the patient presented generalized jaundice without hepatomegaly or abdominal sensitivity.

The initial laboratory screening revealed the following: Aspartate aminotransferase (AST) 246 IU/L, alanine aminotransferase (ALT) 476 IU/L, total bilirubin at 354 mg/l, direct bilirubin: 278 mg/l, alkaline phosphatase (ALP) at 812 IU/l and γ-glutamyl transferase (GGT) at 465 IU / l with a Prothrombin Ratio at 82%. Electrolytes and renal function were normal. Chest x-ray and electrocardiogram (ECG) were reported to be normal.

The morphological assessment, including an abdominal ultrasound, an abdominal CT scan and Magnetic resonance cholangiopancreatography, did not reveal any abnormality of the liver parenchyma or the biliary tree, moreover it showed a magma of retroperitoneal lymphadenopathy (Figure 1).

In this context, the main causes of cholestasis were sought and excluded by Negative or normal tests, namely Serologies for hepatitis A, B, C, and E viruses, HIV, Epstein Barr Virus (EBV), Cytomegalovirus (CMV), herpes simplex virus 1 and 2 (HSV) and immunological analysis (Anti-nuclear, anti-mitochondrial, anti-liver kidney microsome, and anti-smooth muscle antibodies). Furthermore, thyroid hormone values were within the reference values, as well as ferritin, serum and urine copper levels, ceruloplasmin, alpha1 antitrypsin, anti-transglutaminase antibodies, electrophoresis of proteins, and immunoglobulin quantitation.

In view of the normality of the initial etiological cholestasis assessments, a liver biopsy was performed which revealed intrahepatic cholestasis with

canalicular stasis through an inflammatory infiltrate without any detectable specific lesion (Figure 2).

Given the presence of retroperitoneal lymphadenopathy, a scanno-guided biopsy was performed, which concluded to the diagnosis of sclero-nodular Hodgkin lymphoma.

A pet-scanner was carried out as part of the lymphoma extension assessment confirming the absence of lymphomatous infiltration of the liver.

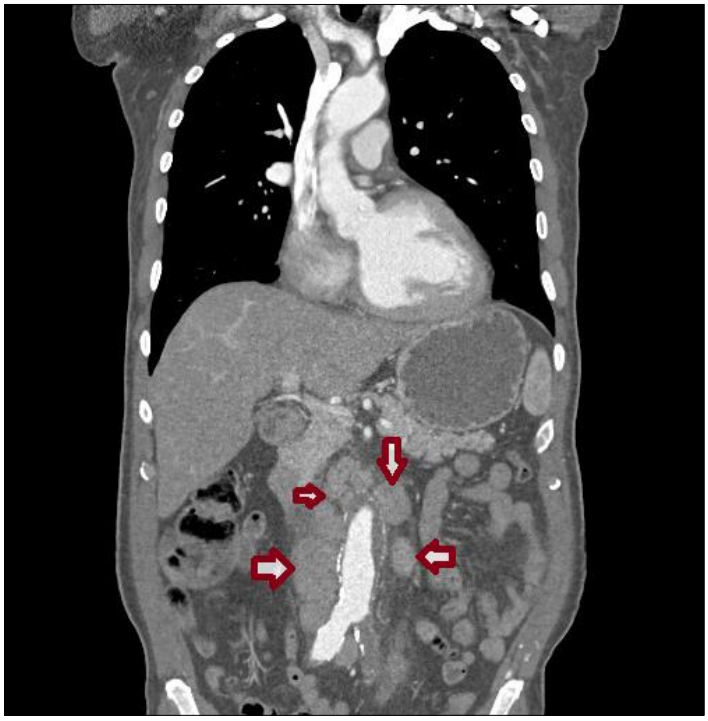

Figure 1: abdominal CT scan did not reveal any abnormality of the liver parenchyma of the biliary tree, moreover it showed a magma of retroperitoneal lymphadenopathy.

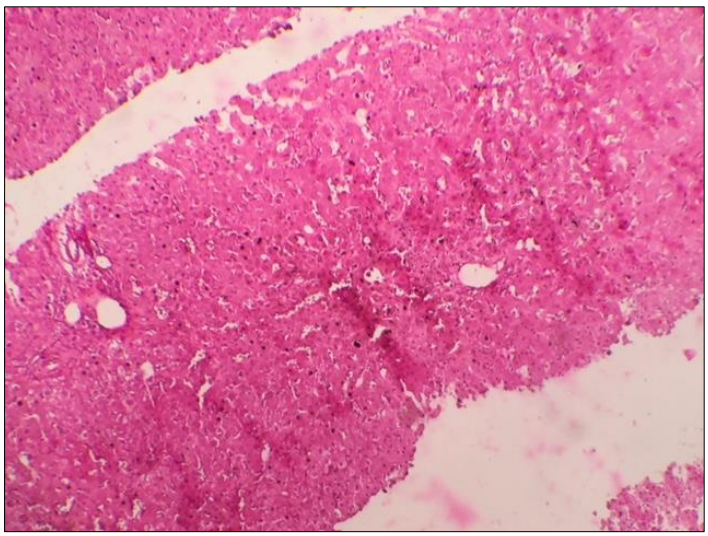

Figure 2: intrahepatic cholestasis with canalicular stasis through an inflammatory infiltrate without any detectable specific lesion (HE x 10).

DISCUSSION

Cholestasis in HL is rare, and the most common pathophysiological mechanism responsible, is hepatic parenchymal or epithelial infiltration by the lymphoma cells [1]. Sometimes it is due either to obstruction of the extrahepatic bile ducts by lymphadenopathy, or more rarely it is explained by another underlying condition, other than lymphoma, such as infectious hepatitis, liver disease, or drug toxicities [1, 2, 3, 4].

Exceptionally, this cholestasis is part of a Paraneoplastic Syndrom linked to Hodgkin lymphoma,

divided into 2 subgroups according to the histopathological results of the liver biopsy, which are Vanishing bile duct syndrom: described for the first time by Hubscher in 1993, and defined by the destruction of the intrahepatic bile ducts involving more than 50% of the portal spaces due to an autoimmune mechanism leading to ductopenia. [5, 6].

And Idiopathic intrahepatic cholestasis, which corresponds to our case, this phenomenon was described for the first time in 1962 by BOURONCLE [1], and the exact pathogenesis of cholestasis in this case is not clear, however the likely mechanisms would be the secretion of a cholestatic cytokine or the development of hormones such as androgens or 17-alkyl estrogen derivatives from tumor cells [7]. The anatomopathological study in this case most often shows a canalicular stasis by mixed inflammatory infiltrates [8].

This phenomenon represents both a diagnostic problem because this diagnosis must be retained after eliminating all other causes of cholestasis, and a therapeutic problem because the decision and the moment for treatment puts the practitioner in a balance between the hepatotoxicity of chemotherapy and the progression of the lymphomatous disease.

Declaration of Interests

The authors declare that they have no conflicts of interest in relation to this article.

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