

A 20-Year-Old Woman with Stenosis and Dilations of the Biliary Tree

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Case Report

A 20-year-old woman presented with epigastric pain and jaundice. Physical examination revealed mild icteric sclera and normal liver and spleen. Biochemical tests were as follows: normal blood count, alanine aminotransferase 191 U/l, gamma-glutamyltransferase 149 U/l, alkaline phosphatase 252 U/l, total bilirubin 5.6 mg/dl (5.0 conjugated), normal renal function tests. Viral markers for hepatitis A, B, C, Epstein Barr, HIV, CMV were negative, and the immunological profile was normal. She reported occasional alcohol consumption and sporadic cigarette smoking habits. She was not taking medication, but reported an assumption of Levonorgestrel (750 mcg) after a sexual intercourse 2 weeks before. Abdominal ultrasound showed dilatation of intrahepatic biliary ducts; a nuclear magnetic resonance cholangiography (MRC) showed enlarged liver with focal inflammatory areas, splenomegaly, intrahepatic biliary tree enlargement with irregular profiles and marked stenosis alternated to dilatations and cystis; the common hepatic duct was mild dilated without stenosis, compatible with primary sclerosing cholangitis (PSC) (Figure 1). An inflammatory bowel disease was ruled out by a colonoscopy. The patient was treated with standard dose of ursodeoxycholic acid, followed by a gradual normalization of liver



Figure 1: Maximum intensity projection image showing diffuse dilatation and irregularity of the intrahepatic ducts in the left and right liver lobe, with multiple marked saccular dilatation and segmental narrowing; mild segmental dilatation of the common hepatic duct is also visible. No intraluminal filling defects are present.

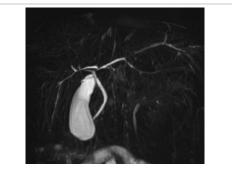


Figure 2: Maximum intensity projection image showing remarkable resolution of the typical features of acute cholangitis, particularly of the dilatation.

function tests. A MRC performed after 2 months revealed a normal biliary tree with disappearance of the biliary changes (Figure 2).

The total disappearance of the cholangiographic abnormalities suggests a secondary form of sclerosing cholangitis (SSC). SSC is a rare condition determined by a variety of causes including post-traumatic or surgery conditions, ischemia, cholelithiasis/choledocolithiasis, haematologic conditions (eosinophilic and mast-cell abnormalities), hepatic inflammatory pseudotumor, AIDS, infectious conditions (recurrent pyogenic cholangitis), infiltrative disorders, metastasis, congenital hepatic diseases [1]. Among the SSC only the eosinophilic cholangitis has been reported to have a complete regression of the biliary abnormalities, particularly after steroid treatment [2].

The unique risk factor for hepatic damage in the patient was the consumption of Levonorgestrel. Drug-induced liver injury may be characterized by a cholestatic pattern with biochemical alterations in absence of morphological abnormalities of the biliary tree. Recently, 8 cases of drug-induced liver disease with biliary abnormalities at the MRC have been reported [3].

Anecdotal cases of low dose levonorgestrel/ethinylestradiol hepatotoxicity have been reported in young females taking pill as anti-contraceptive [4,5]. In these cases liver biopsy showed sinusoidal dilatation or focal necrosis in peri- and centrolobular area, and liver damage resolved after pill discontinuation. In 2 cases endoscopic retrograde cholangiogram showed normal biliary tree.

These evidences suggest a possible role of emergency contraception with Levonorgestrel in cholestatic liver injury. Usually a cholestatic toxicity due to a drug-induced liver injury is characterized by biochemical alterations in absence of morphological abnormalities. Our case report is relevant because for the first time we pinpointed the link between Levonorgestrel and biliary tree alterations which had a complete regression after the drug discontinuation.

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