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LETTER TO THE EDITOR

Primary biliary cirrhosis developing in a patient with Crohn's disease during the course of infliximab treatment: The first case in the literature

Dear Sir,

Primary biliary cirrhosis (PBC), the association of this entity with ulcerative colitis rarely has been reported and it was speculated whether the two diseases could share a common immunologic basis.[1] However, PBC with Crohn's disease (CD) was only reported once, accepting it as an extraintestinal manifestation (EIM) of CD rather than an adverse event of IBD treatment (Tx).[2] Herein, we report the first case of PBC developing under Infliximab (IFX) Tx in a patient with CD.

A 44-yr-old female patient was followed up in our IBD outpatient clinic with ileocolonic CD for 14 years. After the disease onset she had regularly used 5-ASA compounds for the first four years but then stopped her Tx for ten years on her own. Then combo Tx with azathioprine (AZA) and IFX was started due to inflammatory stricture in the terminal ileum. Prior to combo Tx her liver function tests, all immunoglobulin (Ig) levels were within normal limits and antinuclear antibody (ANA), and serologic tests for common hepatitis virus including A, B, and C were all negative and her initial physical examination was normal and she had neither jaundice, liver–spleen enlargement, nor any stigmata of chronic liver disease.

After starting the Tx serum ALP and GGT levels gradually elevated while serum AST, ALT and bilirubin levels remained normal. When the first slight increase was noted before the third IFX infusion, AZA immediately was stopped suspecting toxic cholestasis but IFX Tx continued. However, after the fourth infusion ALP and GGT levels increased to four-times of the upper normal limit (Fig. 1) and it was found that serum antimitochondrial antibody (AMA) showed a high-titer (1/1280) positivity and serum IgM was 846 mg/dl (normal, 40–320) in the presence of a normal magnetic resonance cholangiopancreatography. Therefore, a percutaneous liver biopsy was performed disclosing features consistent with PBC (Fig. 2). IFX Tx was stopped and liver enzymes, AMA and IgM levels returned to normal levels within one month. Afterwards the patient underwent a right hemicolectomy and immediately was put on AZA to prevent disease recurrence. She has no further problems under AZA during six months close postoperative follow up period.

To our best knowledge there is only one reported patient with CD developing PBS, actually thought to be an EIM of CD by the authors.[2] In our case PBS manifested after the start of combo Tx with AZA and IFX and immediately subsided after the cessation of Tx with normalization of liver enzymes and AMA and IgM titers. The problem-free follow up under solo AZA lets us suggest that the offending drug was IFX and this

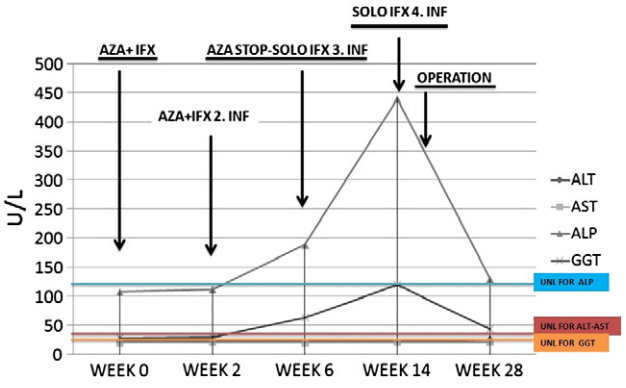

Figure 1 The course of liver enzymes under IFX + AZA Tx (ULN: upper limit of normal).

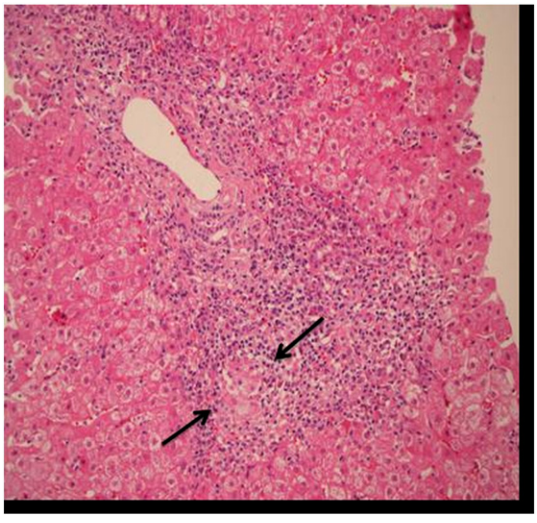

Figure 2 Inflammatory infiltrates in portal areas with irregular bile duct epithelium and granuloma formation (arrowheads) (HEX200).

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makes the case an interesting one as this is the first PBS case developing under IFX Tx.

Conflict of interest

All authors declared no conflict of interest.

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