

Rare Association Of Budd Chiari Syndrome And Peliosis Hepatis : Could Oral Contraception Be The Cause?

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ABSTRACT

Oral contraceptive pills are widely used for birth control, but they have been associated with various hepatic disorders. This article presents a unique case of the simultaneous occurrence of Budd-Chiari syndrome and peliosis hepatis, which is likely attributed to long-term use of oral contraception. The case involves a 38-year-old woman with a history of oral contraceptive use since adolescence who initially consulted for abdominal ascites.

INTRODUCTION

Oral contraceptive pills are the most commonly used means of birth control in women. Several studies have demonstrated the association between the use of oral contraceptives and several hepatic disorders including: Cholestasis, hepatic neoplasms, as well as vascular pathologies such as portal vein thrombosis, Budd Chiari syndrome and peliosis hepatis [1].

We will present in this article an exceptional association of Budd Chiari syndrome and peliosis hepatis whose most probable origin is the prolonged use of oral contraception.

PRESENTATION OF THE CASE

We present here the case of a 38-year-old patient, married and mother of 3 children. She had taken oral contraception since the age of 13 years, without risk factors of chronic liver disease, nor personal or family history of venous thrombosis. She consulted a hepato-gastroenterologist 8 months ago for abdominal pain and progressive abdominal distension, in whom the clinical examination showed the presence of ascites.

A blood test was performed, the blood count came back without any particularities, notably no anemia (Hemoglobin at 11.5 g/dl) and a correct platelet count at 170.000 elements/mm³. The prothrombin level was slightly decreased to 67% with a correct albumin level.

There was no cytolysis or cholestasis. An exploratory puncture of the ascites fluid showed a protein level of 32g/l.

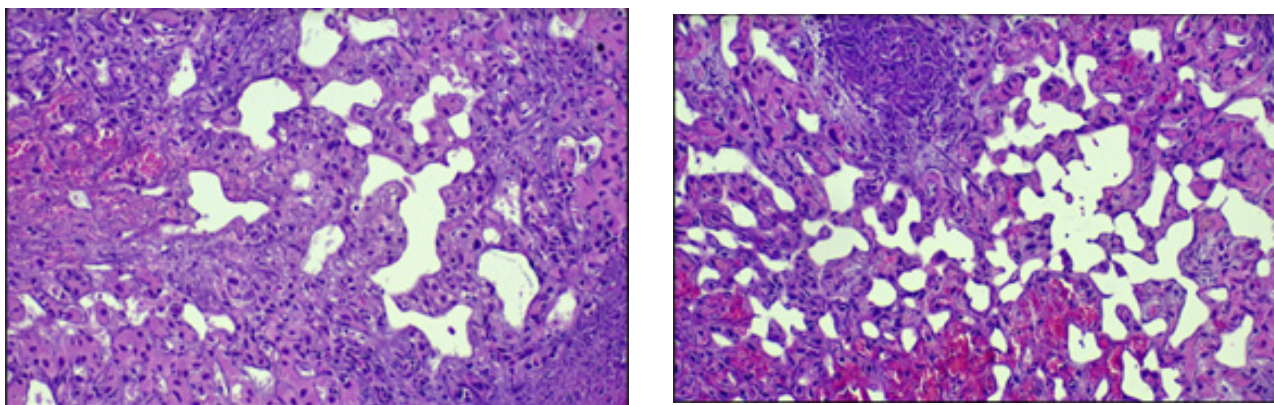
Abdominal ultrasound showed a heterogeneous liver with irregular contours, in favor of a chronic liver disease, as well as an absence of visualization of the three suprahepatic veins, with a permeable portal trunk dilated to 14 cm and a homogeneous splenomegaly. The gallbladder was multi-lithiasic. The diagnosis of Budd Chiari syndrome was confirmed by angioscan. On the other hand, hepatitis B and C serologies came back negative.

The patient underwent a liver biopsy during cholecystectomy which showed chronic hepatitis associated with a marked hepatic peliosis.

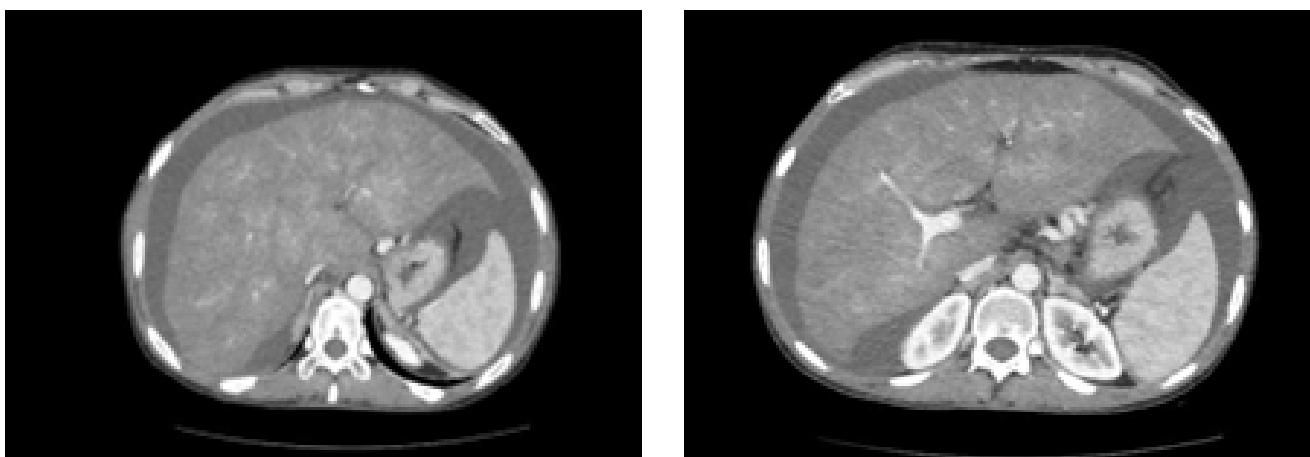
A thrombophilia test was performed, which showed the absence of the JAK2V617F mutation, as well as the factor 5 Leiden mutation, the absence of protein C and S deficiency. An antiphospholipid syndrome was also ruled out as well as paroxysmal nocturnal hemoglobinuria.

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Figure 1 and 2 : Extensive sinusoidal dilatation in the liver lobules, bordered by regular epithelial cells and hemorrhagic remodeling compatible with peliosis hepatis.



Figures 3 and 4: Enhancement of the hepatic parenchyma in a mosaic pattern associated with vascular congestion and the presence of sub-centimeter regenerative hepatic nodules, with the absence of visualization of the suprahepatic veins related to Budd-Chiari syndrome.



DISCUSSION

Budd-Chiari syndrome is defined as obstructed hepatic venous outflow due to occlusion of the hepatic veins or inferior vena cava. The etiopathogenic factors are dominated by myeloproliferative syndromes, which are present in 40 to 50% of cases. Other etiologies are represented by hereditary prothrombotic conditions, firstly the factor V Leiden mutation, followed by protein C or S deficiencies, as well as acquired diseases such as antiphospholipid syndrome and paroxysmal nocturnal hemoglobinuria [2].

Oral contraception is reported with a variable frequency depending on the series, and seems to be more particularly associated with thrombosis of the suprahepatic veins compared to thrombosis of the inferior vena cava. A study by Valla et al. Indicated that oral contraception users are more than twice as likely to develop hepatic vein thrombosis compared with nonusers [relative risk (RR), 2.37; 95% confidence interval (CI): 1.05-5.34, p less than 0.02] [3].

Although, in the most recent multicenter series, and after the discovery of several prothrombotic conditions, the role of contraceptives seems to be more of an enhancer of an underlying thrombophilia, triggering the development of hepatic vein thrombosis.

Therefore, even in cases where oral contraception appears to be strongly incriminated as the only obvious thrombogenic risk factor, it is critical to exclude other factors, including hereditary thrombophilia and latent or overt myeloproliferative disease. On the other hand, peliosis is defined by the presence of multiple blood-filled cystic cavities which may be seen in the liver, spleen, lymph nodes or other organs. Peliosis hepatis is a rare disorder which may be associated with, or caused by, various diseases, drugs (including oral contraceptives but also Azathioprine, 6-thioguanine and oxaliplatin.), and infections

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(Such as Bartonella infection and tuberculosis) [4,5]. Unlike azathioprine and the 6-Thioguanines which lead to hepatic peliosis by causing a sinusoidal endothelial cell injury, the mechanism by which oral contraceptives lead to peliosis hepatis remains uncertain [6].

Peliosis hepatis can be detected incidentally at hepatic imaging or laparoscopy in asymptomatic individuals, but intraperitoneal hemorrhage can occur as a catastrophic presentation. Cases with hepatomegaly, portal hypertension, and liver failure have also been reported [7]. The diagnosis when confirmed by liver biopsy can show: Hemorrhagic parenchymal necrosis with blood cavities neither lined by sinusoids/fibrosis or aneurysmal dilatation of central vein with cavities lined by endothelium and fibrosis [8]. The lesions show regression after interruption of usage with incomplete response, in some cases progressing to cirrhosis, which is the case of our patient.

The particularity of our case lies in the fact that oral contraception seems to be the only factor explaining the rare association between Budd Chiari syndrome and peliosis hepatis.

CONCLUSION

This case highlights the potential association between long-term use of oral contraception and the development of both Budd-Chiari syndrome and peliosis hepatis. While oral contraceptives are commonly used and generally considered safe, it is essential to recognize their potential hepatotoxic effects and the increased risk of hepatic vein thrombosis, particularly in susceptible individuals. Therefore, clinicians should remain vigilant in monitoring patients using oral contraceptives for any signs or symptoms of hepatic dysfunction and consider alternative contraceptive methods in individuals at higher risk.

Conflict of interests : The author reports no conflict of interest.

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