

Bilateral Pleural Effusion: A Rare Case Report

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Abstract

A 24-year-old female presented with complaints of distension of abdomen, lower limb swelling and shortness of breath. On examination, she had bilateral pedal oedema, tender mild hepatomegaly, bilateral pleural effusion and ascites. She had received treatment on lines of chronic liver disease, congestive heart failure but did not manifest any improvement. Laboratory investigations including haemogram, echocardiography and liver function testing were inconclusive. Ultrasonography and computed tomography of abdomen revealed obstruction at the junction of inferior vena cava and hepatic vein with pre-stenotic dilatation confirming the diagnosis of Budd-Chiari syndrome. She was treated with balloon dilatation from right femoral vein and the patient showed marked recovery with decrease in ascites and bilateral pleural effusion and improvement in dyspnoea and leg swelling. [Indian J Chest Dis Allied Sci 2015;57:243-245]

Key words: Inferior vena cava obstruction, Ascites, Pleural effusion, CHF, Budd-Chiari syndrome, Balloon dilatation.

Introduction

Budd-Chiari syndrome is an uncommon condition induced by thrombotic or non-thrombotic obstruction of hepatic venous outflow; obstruction can be at any level starting from hepatic venules to inferior vena cava (IVC). Classically, the triad of abdominal pain, hepatomegaly and ascites characterise this syndrome. In the Indian scenario, 50% to 60% cases of Budd-Chiari syndrome are idiopathic.^{1,2} Haematological disorders, inherited thrombotic diathesis, pregnancy and post-partum period, oral contraceptive pills, chronic infections, chronic inflammatory disorders, tumours, alpha-1 antitrypsin deficiency, trauma and anti-cancer drugs are common causes of Budd-Chiari syndrome. We report an atypical presentation of Budd-Chiari syndrome.

Case Report

A 24-year-old female presented with complaints of distension of abdomen from two-and-a-half years, lower limb swelling from eight months and shortness of breath from eight days. The patient was apparently well till two-and-a-half years ago when she noted distension of abdomen. Ultrasonography of the abdomen was done and repeated after three months which showed mild hepatomegaly with generalised

depressed echotexture with non-obstructive hepatocellular changes and moderate ascites. She was empirically treated as a case of chronic liver disease with portal hypertension; her ascites was repeatedly tapped and she had received diuretics and salt restriction, but there was no response. Computed tomography (CT) of the abdomen was done after six months which showed moderate ascites with bilateral effusion, enlarged right ovary, left ovary was normal, so a right oophorectomy was done and a chocolate cyst removed from her right ovary, but the patient did not improve. Because of bilateral lower limb swelling and tender hepatomegaly, she was also treated as a case of congestive heart failure (CHF), but again, there was no relief. Her past medical history was insignificant. Physical examination revealed decreased breath sounds in both infrascapular areas (right-side > left-side) and per abdominal examination showed abdominal distension with fluid thrill and tender hepatomegaly.

Laboratory testing revealed haemoglobin 10 g/dL; total leucocyte count 8100/mm³; with a differential count of polymorphs 78%, lymphocytes 22%; erythrocyte sedimentation rate was 30 mm at the end of the first hour; total serum proteins 4 g/dL; serum albumin 1.5 g/dL, serum aspartate aminotransferase (AST) 68 IU, serum alanine aminotransferase (ALT) 51 IU,

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serum alkaline phosphatase 226 IU; blood urea nitrogen 12 mg/dL; serum creatinine 0.6 mg/dL. Urine routine and microscopic examination and coagulation profile were normal. Ascitic fluid examination yielded protein 0.8 g/dL; sugar 140 mg/dL; cell counts 50 cells /mm³ (all lymphocytes); ascitic fluid adenosine deaminase levels were 13.7 IU/L and pyogenic culture was sterile. Chest radiograph showed bilateral pleural effusion (Figure 1). Aspirate from right pleural space also yielded a transudative pleural fluid. Electrocardiogram (ECG) was within normal limits. Echocardiography was normal; no regional wall motion abnormalities or valvular lesions were observed and left ventricular ejection fraction was 62%.

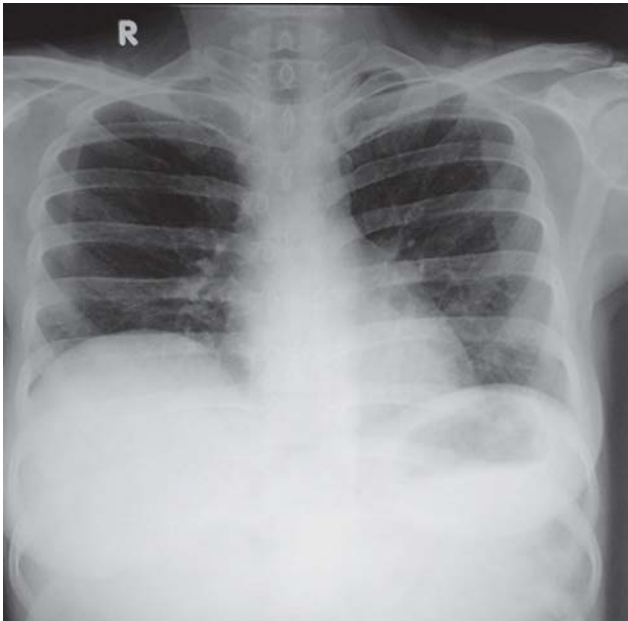


Figure 1. Chest radiograph (postero-anterior view) showing bilateral pleural effusion.

Ultrasonography of abdomen showed obstruction at the junction of IVC and hepatic vein with pre-stenotic dilatation, loss of respiratory phase variation of IVC on colour Doppler, congestive hepatomegaly with coarse echotexture and moderate ascites. Contrast enhanced computed tomography (CECT) of upper abdomen and lower chest revealed hepatomegaly with nodular spotty pattern of contrast enhancement in liver with enlarged caudate lobe with non-visualisation of hepatic veins and small segment of luminal narrowing in IVC (Figure 2) with marked ascites and loculated right pleural effusion. She was diagnosed to have Budd-Chiari syndrome. Laboratory testing for protein C, protein S, factor V Leiden, anticardiolipin and antiphospholipid antibodies and antithrombin III levels were found to be within normal range. Serological testing for human immunodeficiency virus (HIV), hepatitis B and C viruses was negative. Serological markers for amoebiasis and echinococcus were also negative. Following an urgent Cardiology consultation,



Figure 2. Contrast enhanced computed tomography of the abdomen showing inferior vena cava narrowing.

IVC balloon dilatation was done through right femoral vein with balloon MP1 (12*4, 16*4, INOVE 24) with omnipaque dye. The procedure was successful (Figure 3). The patient started improving, ascites reduced along with decrease in pleural effusions and she reported reduction in dyspnoea and leg swelling. The patient

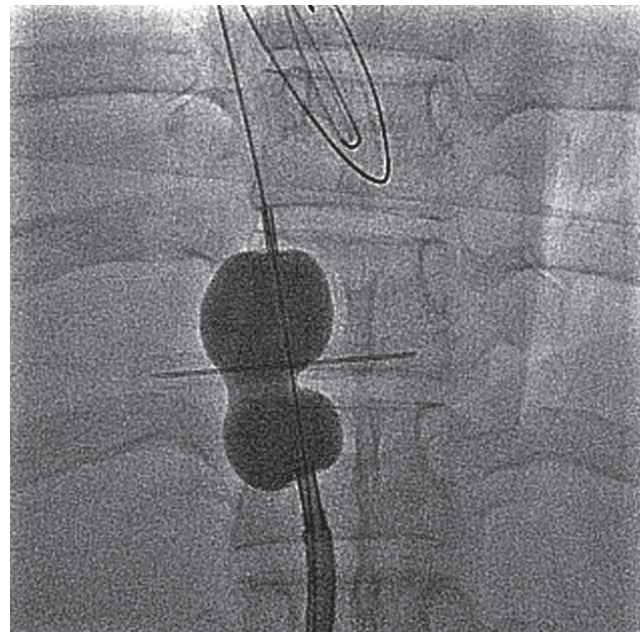


Figure 3. Inferior vena cava balloon dilatation showing balloon inflating the narrowed inferior vena cava.

was discharged on short-term antibiotics and diuretics. She has been followed-up for one year. There has been no recurrence of ascites or pleural effusion.

Discussion

Budd-Chiari syndrome was described by Budd in 1845³ and Chiari added the first pathologic description of a liver with obliterating endophlebitis of the hepatic veins in 1899.⁴ Budd-Chiari syndrome is an uncommon condition induced by thrombotic or non-thrombotic obstruction of hepatic venous outflow; obstruction can be at any level starting from hepatic venules to IVC. Hepatomegaly, ascites and abdominal pain make the classic triad of Budd-Chiari syndrome. A high index of suspicion is necessary to make the diagnosis. The aetio-pathology can be described as obstruction of intra-hepatic veins leading to congestive hepatopathy and hepatocellular injury resulting in microvascular ischaemia.^{5,6} There is no gender predilection, the disease usually occurs in third and fourth decades of life and is potentially fatal if untreated. Medical therapy alone is associated with a high 2-year mortality rate ranging from 80%-85%.⁷ There are four clinical variants, the acute and the subacute form, the chronic form which is most common and the fulminant form.⁵ Diagnosis is usually confirmed by ultrasonography, CT with pulse sequencing and hepatic venography.

In the present case, ultrasonography alone was diagnostic of the syndrome. In another series,⁸ 80% of cases could be diagnosed by ultrasonography with Doppler study; remaining 20% required CT/ magnetic resonance angiography for the diagnosis.⁸

Mechanism of Budd-Chiari syndrome in our case was obstruction at the junction of IVC and hepatic vein, which is consistent with the Indian studies on Budd-Chiari syndrome.^{1,2,6,9} In the western countries, hepatic vein thrombosis remains responsible for majority of Budd-Chiari syndrome; IVC obstruction is rarely found.⁸ The case presented here is unique because of many reasons. Her initial presentation was not typical; she started with distension of abdomen and lower limb swelling but without any pain abdomen or tender hepatomegaly.

Depending upon the severity of IVC obstruction, many modalities have been reported in the literature,

namely, IVC balloon angiography, IVC stenting, combined IVC and hepatic vein stenting, isolated hepatic vein stenting, transjugular intra-hepatic portosystemic shunt (TIPS) and IVC stenting with TIPS.⁸ Our patient responded very well to IVC balloon angiography alone.

Though the presentation of Budd-Chiari syndrome with unresolving ascites with bilateral pleural effusion and pedal oedema may be rare, a high degree of clinical suspicion along with good imaging techniques are required to confirm the diagnosis. With chronic form of presentation being more common, it may take few years for a disorder to be confirmed on radiological grounds. The combined obstruction of hepatic and IVC, though an uncommon site of obstruction should be kept in mind in patients with ascites, bilateral pedal oedema and pleural effusion.

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