

## Case report

# A Rare case report of HEPATIC VENOUS OUTFLOW TRACT OBSTRUCTION in patient with ULCERATIVE COLITIS

### ABSTRACT:

We report a case of 23 year old female diagnosed as Hepatic Venous Outflow tract obstruction (HVOTO) with ulcerative colitis (UC) with CMV Colitis. HVOTO with UC is a very rare entity, to the best of our knowledge there have been only few published cases reports of HVOTO with UC. Our patient presented with fever, bloody stools with tenesmus, colicky abdominal pain from last 6 months, and pedal edema with abdominal distention from last 2 months. Sigmoidoscopy and biopsy was done on presentation which was suggestive of active ulcerative colitis with CMV colitis. Color Doppler of spleno-portal axis was done suggestive of HVOTO. Patient was started on Ganciclovir, Mesalamine, anticoagulants and was subjected to balloon angioplasty after which anticoagulants were continued. Patient is being followed up and is doing well.

*KEYWORDS: Ulcerative Colitis, Hepatic Venous Outflow Tract Obstruction, CMV Colitis, Ganciclovir*

### INTRODUCTION:

Patients with inflammatory bowel disease (IBD) are at increased risk for thrombo-embolic complications<sup>1</sup>, but hepatic vein thrombosis has been reported as a rare extra intestinal complication of UC<sup>2</sup>. Hence we report a case of a patient with UC with CMV colitis, complicated by the development HVOTO.

### CASE REPORT:

Our patient was a 23 years old female presented with complaints of 4-5 episode per day of semisolid stools mixed with blood associated with tenesmus, colicky abdominal pain and fever for 6 months. She had complaints of abdominal distention with bilateral lower limb swelling since 2 months which was not associated with jaundice, haemetemesis or malena. The patient was not taking any

medications. On clinical examination, BP was 110/68 mm Hg, Pulse rate was 88/min, tender hepatomegaly, splenomegaly and shifting dullness were present along with dilated veins over anterior abdominal wall and flanks (Figure No.1). Other systemic findings were unremarkable. Haematological and Biochemical investigations of the patient are mentioned in Table No.1. On Sigmoidoscopy there was patchy loss of vascularity with increased friability and granularity, overlying superficial and deep ulcers with spots of coagulated blood were present; Baron score 2, Ulcerative Colitis Endoscopic Index of Severity– 5/8 suggestive of IBD-UC (Figure no. 2). On biopsy, there were changes consistent with ulcerative colitis with cryptitis and crypt abscess formation without dysplasia or granuloma. Few cells were showing nucleomegaly with nuclear inclusions (Figure No.3). CMV DNA was detected, hence the biopsy picture was suggestive of Acute Ulcerative colitis with features of CMV colitis. The ascitic fluid analysis was done suggestive of high Serum Albumin Ascitic fluid Gradient (SAAG) and low protein indicating portal hypertension as the etiology. She was subjected to Color Doppler of hepatoportal system which showed ostial narrowing of all the hepatic veins suggesting Hepatic venous outflow tract obstruction, portal hypertension with liver parenchymal disease and multiple nodules? Dysplastic ? regenerative. She was subjected to Triphasic computed tomography of abdomen which showed short segment narrowing of Middle hepatic vein and Right hepatic vein with left hepatic vein ostial narrowing suggestive of HVOTO with features of Liver parenchymal disease with portal hypertension and multiple nodules ?dysplastic ?regenerative (Figure No. 4). Alpha fetoprotein was normal. CT guided targeted biopsy of Hepatic nodules was done with biopsy report suggestive of high grade dysplastic nodules. Upper GI Endoscopy showed features of early portal hypertension. For ruling out other causes of hypercoagulability like Protein C and S deficiency, AT III deficiency, Factor V leiden mutation, Prothrombin Gene mutation G20210A, Antinuclear antibodies, Antiphospholipid antibodies investigations were sent which turned out to be negative. As mentioned earlier, our patient was not on any oral contraceptive medication. After admission she was started on Mesalamine and Ganciclovir, her stools frequency reduced to 1-2 episodes per day without blood. Patient was also started on diuretics and anticoagulation. Patient was subjected to ostial dilatation of Right Hepatic Vein and Middle Hepatic Vein via balloon angioplasty and anticoagulation was continued after procedure.

## DISCUSSION:

Hepatic Vein thrombosis is a rare extra intestinal manifestation associated with IBD. Worldwide only few cases have been reported. Patients with UC are associated with increased risk for venous thromboembolism (VTE) at baseline but the risk is eight times higher during a flare up<sup>3</sup>. UC is also associated with an increased risk of arterial thromboembolic events<sup>4</sup>. The risk of VTE development among IBD patients is positively associated with both disease extent and activity. VTE in IBD patients occurs earlier in life than in those without IBD<sup>5 6</sup>. These and other findings support the classification of IBD as an independent risk factor for the development of VTE. The Acquisition of non heritable risk factors for thromboembolic disease among IBD patients particularly during acute flare ups is likely contributory. One study also suggested IBD patients remain at higher risk of venous thromboembolism even after proctocolectomy<sup>7</sup>. IBD patients have abnormalities in coagulation<sup>8 9 10</sup>, Platelet function<sup>11 12</sup>, fibrinolysis<sup>13</sup>, endothelial dysfunction<sup>14</sup> and active inflammatory cascade<sup>15</sup>. Cytokines such as Il-1, IL -6, and Anti TNF alpha remain at a higher function during the course of disease<sup>16</sup>. These cytokines function as proinflammatory substances and increase the risk of hypercoagulability. Underlying hypercoagulable disorders can meld with IBD hypercoagulable state. However the hypercoagulability profile in our patient was negative. HVOTO can present in acute, subacute and chronic form. The diagnosis of HVOTO can be

made in patients presenting with abdominal pain, ascites, dilated veins and tender hepatomegaly or with other findings raising a high level of suspicion in the clinician. The diagnostic modalities that have been found to be most helpful are Doppler ultrasound<sup>17</sup> and Computed Tomography<sup>18</sup>. Magnetic Resonance Angiography has been shown in a few studies to be more accurate in delineating the hepatic vasculature to more precisely define the location of the obstruction<sup>19</sup>. The gold standard for diagnosis is hepatic venography but it is more invasive and is typically performed when less invasive methods of evaluation are equivocal or negative. Therapeutic options for HVOTO with IBD are varied and depends on clinical presentation. Anticoagulation should be initiated immediately and continued for life unless contraindicated. Our patient underwent ostial dilatation of Right Hepatic Vein and Middle Hepatic Vein via Balloon angioplasty and was kept on anticoagulants. Patient is being followed up and is doing well. No untoward side effect of anticoagulants were noticed till now.

## **CONCLUSION:**

IBD-UC patients can have varied range of intestinal and extra-intestinal manifestations. Patients with IBD are at increased risk of venous and arterial thrombosis. Hepatic vein or inferior vena cava thrombosis is a rare extra-intestinal complication of ulcerative colitis. There should be a high level of suspicion for HVOTO in patients with IBD presenting with ascites, dilated veins and tender hepatomegaly.

**TABLES:****Table No. 1 Haematological and Biochemical investigations of the patient :**

Haemoglobin	10.6 g/dL (14-16 g/dL)
TLC	5870/ cu.mm(4000-11000 / cu.mm)
PLT	0.55 L/cu.mm (1.5-4.5 L/cu.mm)
CRP	35.4 mg/L(<3mg/L)
KIDNEY FUNCTION TESTS	Urea – 10mg/dL (20-40 mg/dL) Creatinine – 0.9mg/dL (0.6-1.2 mg/dL) Sodium – 132 mEq/L(135-145 mEq/L) Potassium –3.4 mEq/L(3.5-5.5mEq/L)
LIVER FUNCTION TESTS	SGPT – 13 IU/L (5-37 IU/L) SGOT – 49 IU/L (7-40 IU/L) ALP – 359 IU/L (40-150 IU/L) Total Bilirubin – 1.9 mg/dL (0.2-1.3 mg/dL) Direct Bilirubin –1.0 mg/dL (0-0.3 mg/dL)
ALBUMIN	2.1g/dL
GLOBULIN	2.9 g/dL
PT	16.8s (11-15)
INR	1.12
HIV/HBsAG/ANTI-HCV	NR/NR/NR
Ascitic Fluid Analysis	TP – 1.48g/dL ALB – 0.79 g/dL TLC – 80 cells/cu.mm DLC – N -14/L -79 SAAG – 1.31

Figure No.1 Dilated Veins present on anterior abdominal wall.



Figure No. 2 Sigmoidoscopy finding of patchy loss of vascularity with increased friability and granularity, overlying superficial and deep ulcers with spots of coagulated blood; Baron score 2, Ulcerative Colitis Endoscopic Index of Severity– 5/8 suggestive of IBD-UC

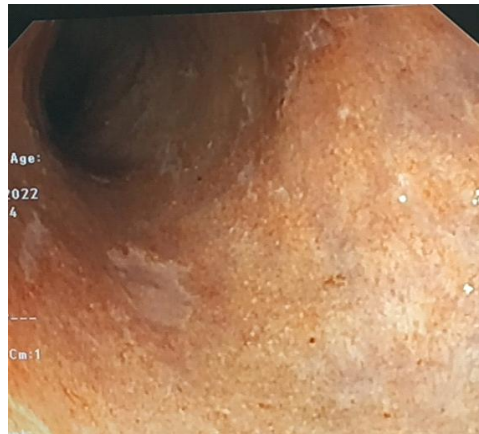


Figure No. 3 Histopathological picture of UC with few cells showing nucleomegaly and nuclear inclusions suggestive of CMV colitis.

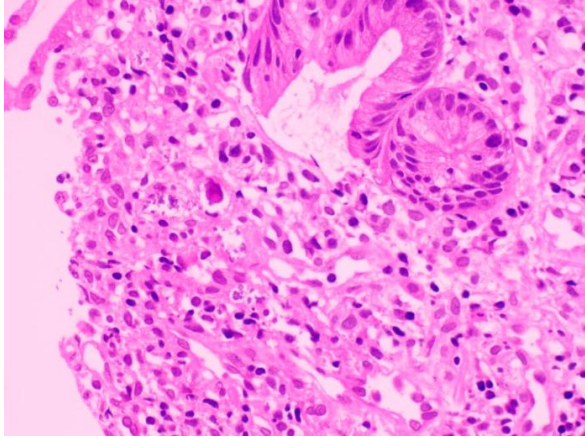
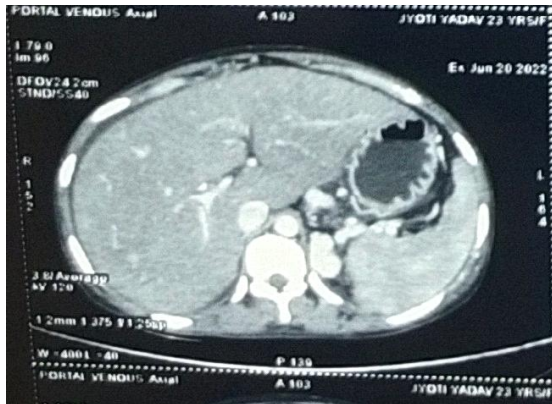


Figure No. 4 TPCT image suggestive of short segment narrowing of Middle hepatic vein and Right hepatic vein with left hepatic vein ostial narrowing suggestive of HVOTO with features of Liver parenchymal disease with portal hypertension and multiple nodules ?dysplastic?regenerative.



## CONSENT

All authors declare that written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editorial office/Chief Editor/Editorial Board members of this journal.

## ETHICAL APPROVAL

No ethical approval was required for this manuscript.

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UNDER PEER REVIEW