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Celiac Disease Presenting as Celiac Artery Stenosis and Intra Abdominal Venous Thrombosis - An Unheard Entity

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Abstract Extra intestinal manifestations of celiac disease are seen in about 20% of patients including venous thrombosis but arterial stenosis is not described in the literature. We describe a first-ever case of celiac disease in 26-year-old lady presenting as severe celiac artery stenosis, managed successfully with arterial stenting. She also had portal and superior mesenteric vein thrombosis. Excellent improvement was seen during follow up after treatment with a gluten-free diet, oral anticoagulation, antiplatelet and celiac arterial stenting. Hypercoagulability and thromboembolic manifestation in celiac disease should be kept in mind especially during active disease or acute exacerbation of celiac disease.

Keywords: *venous thrombosis, celiac artery stenosis, villous atrophy, aortography*

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1. Introduction

Celiac disease (CD) is a chronic small intestinal immune-mediated enteropathy precipitated by exposure to dietary gluten in genetically predisposed individuals. [1] It presents with intestinal and extra intestinal manifestations. Many adults present with gastrointestinal symptoms including diarrhoea, steatorrhea, flatulence, and weight loss. Extra intestinal features, including anaemia, osteopenia, neurologic symptoms, and menstrual abnormalities, seen in about 20% of patients, often prove more distressing to the patient than do the gastrointestinal symptoms.

Hematological manifestations like anemia, hemorrhage, thrombocytosis and vascular manifestations like venous thrombosis though rare but known in CD but there is no available literature on arterial stenosis in CD. We describe the first ever reported case of CD presenting as severe celiac artery stenosis, managed successfully with arterial stenting with portal and superior mesenteric vein (SMV) thrombosis.

2. Case Description

A 26-year-old female presented with severe persistent upper abdominal pain worse after eating and relieved partly by leaning forward, anorexia, vomiting since last one month and chronic diarrhoea, weight loss, growth retardation since early childhood. Primary amenorrhoea and chronic anaemia since childhood was significant past history.

Physical examination revealed severe growth retardation (BMI- 15.92 kg/m^2), height 148 cm, pallor, icterus and soft mildly tender upper abdomen.

Table 1. Blood investigations on admission

Investigation at admission with Reference value	Results
Haemoglobin (g/dl) (13-16)	8
MCV (fl) (80-96)	73
Bilirubin (Total/Direct) (mg/dl) (0.2-1.2 /<0.3)	3/1.5
SGPT (U/L) (5-40)	26
SGOT(U/L) (5-40)	63
ALP (U/L) (20-140)	223
Protein (g/dl) (6-8)	8.8
Albumin (g/dl) (3.5-5)	3.5
PT-INR (1)	1.65
CRP (< 6 mg/L- normal)	49
LDH (U/L)	505

Investigations revealed iron deficiency anaemia, increased C- reactive protein, prothrombin time, LDH with altered liver function tests, normal thyroid-stimulating hormone (Table 1) with positive anti endomysial antibody. Viral markers (hepatitis B and C) negative and autoimmune liver profile was negative with normal IgG level. Ultrasound revealed mild splenomegaly, mildly altered echo texture of liver, portal and SMV thrombosis and changes of portal biliopathy. Gastroscopy revealed small esophageal varices and flat duodenal folds. Diagnosis of CD was made based on clinical features, blood parameters, and positive serology and D2 biopsy features of total villus atrophy. Thrombophilia work up

showed only mildly reduced Protein C, Protein S and antithrombin III .CECT abdomen showed 70% stenosis of celiac artery trunk, SMV and portal vein thrombosis with changes of portal biliopathy .

No relief of abdominal pain even after gluten-free diet and anticoagulation with conventional heparin for 72hrs. Abdominal aortography by Computed tomography revealed ostial 80% stenosis of celiac artery with acute band (Figure 1), same confirmed by conventional angiography and treated with stenting, post stenting, Thrombolysis in Myocardial Infarction (TIMI) Score-3 noted. (Figure 2). Colour doppler study of celiac artery one-week post stenting showed normal flow and velocity in celiac artery. Abdominal pain subsided fully with normal appetite within days after stenting of celiac artery.

Figure 1. Abdominal aortography

She was kept on a gluten-free diet, nutritional supplements, antiplatelet and oral anticoagulation on discharge with regular follow up. On follow up at 6 months, an excellent improvement was seen in the form of no abdominal pain, good weight gain and normalization of haemoglobin (14.7g/dl) and liver function tests.

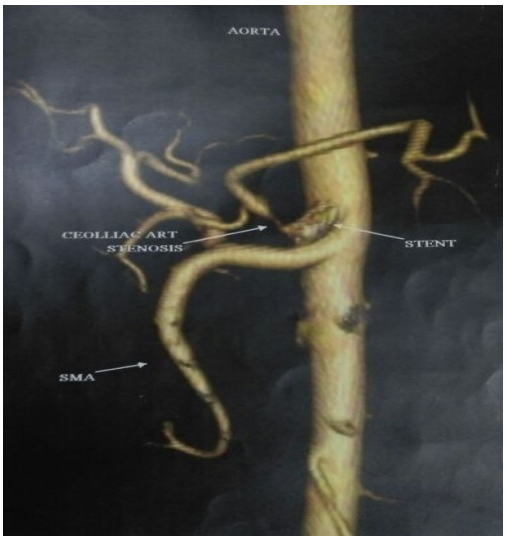

Figure 2. Stent placement for celiac artery stenosis

3. Discussion

In India the incidence of CD is rising & more prevalent in northwest India with female predominance F: M- 1.3 to 2:1. It affects all age groups from infants to old age, 25% of cases diagnosed in patients older than 60 yrs. [2] CD is one of the most commonly missed diagnosis in gastroenterology. Extra intestinal manifestations of CD often result from nutrient malabsorption and can involve virtually all organ systems. A newly explored area of CD is hypercoagulability and the resulting thromboembolic phenomena. There is an increased risk of stroke in adults and children with CD. Thrombophilia, pregnancy loss, deep vein thrombosis, small bowel infarction, atrial fibrillation, Budd-Chiari syndrome, portal and splenic vein thrombosis, and cardiovascular disease have been described. [3] To our knowledge there is no reported case or literature on arterial stenosis in CD.

Few data is providing a causal relation of procoagulation state in the promotion of thromboembolism in CD. In some cases, thromboembolism has been attributed to acquired hyperhomocysteinemia as a consequence of folic acid and vitamin B12 deficiency. [4] Though in our case homocysteine level was normal with negative thrombophilia screening. No risk factors for arterial stenosis and venous thrombosis other than CD was seen in our case explaining the higher risk of thromboembolism in celiac disease.

We report the first case to our knowledge of CD presenting as severe abdominal pain due to celiac artery stenosis, managed successfully with arterial stenting. She also had portal hypertension with portal and SMV thrombosis. Excellent recovery was seen in all clinical and biochemical parameters on gluten-free diet at follow up. She gained 15 Kg weight at one year follow up with normal menses.

No satisfactory explanation could be postulated for celiac artery stenosis in this patient from the available current literature who had undiagnosed CD for about two decades.

In conclusion, hypercoagulability and thromboembolic manifestation in CD should be kept in mind especially during active disease or acute exacerbation of CD.

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