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Case Report

Immune thrombocytopenic purpura as a rare extra-intestinal manifestation in ulcerative colitis: A case report

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ABSTRACT

Immune thrombocytopenic purpura (ITP) is characterized by immune-mediated platelet destruction of platelets. The condition involves the presence of autoantibodies targeting platelet membrane antigens. ITP has been documented in the literature as a rare extra-intestinal manifestation of inflammatory bowel disease (IBD). This case report aims to describe the rare association of ITP as an extra-intestinal manifestation of ulcerative colitis (UC), accompanied by a literature review.

We report the case of a 21-year-old male presenting with acute bloody diarrhea refractory to broad-spectrum antibiotic therapy. Given the patient's significant history of intermittent persistent diarrhea, hematochezia, hematuria, and thrombocytopenia, an evaluation for relapsing IBD was undertaken. Colonoscopic biopsy findings were consistent, with UC showing mild activity, while bone marrow aspiration revealed features indicative of chronic ITP. The patient was managed with oral corticosteroids for the IBD flare and oral mesalamine, resulting in a significant improvement in platelet count. Upon follow-up, with remission of UC, it was accompanied by a complete normalization of the patient's platelet count. ITP has been reported as a rare extra-intestinal manifestation of IBD (UC). This report emphasizes the importance of suspecting IBD in cases of unexplained thrombocytopenia, particularly in compatible clinical settings, to enable timely diagnosis and management of both conditions, ultimately improving patient outcomes.

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INTRODUCTION

Ulcerative colitis (UC) is an inflammatory bowel disease resulting in inflammation and colon and rectum ulcers.[1] The primary symptoms of active disease are abdominal pain and diarrhoea mixed with blood. Other symptoms may include weight loss, fever and anaemia.[2] UC is characterized by remission and recurrence; several extra-intestinal manifestations have been reported. The main extra-intestinal manifestations are arthritis and arthropathy, primary sclerosing cholangitis, metabolic bone disease, uveitis and rarely venous thromboembolism.[3] Immune thrombocytopenic purpura (ITP) is a rare extra-intestinal manifestation of inflammatory bowel disease (IBD) and is more commonly associated with UC.[4] It is an immune-mediated destruction of platelets.[5] Molecular mimicry between the luminal mucosa and platelet membrane antigens leads to immune activation and subsequent platelet destruction in the spleen.[6]

IBD patients are at an increased risk of having another autoimmune disease, and therefore, it becomes even more challenging to distinguish between extra-intestinal manifestation and concomitant autoimmune disease. The association between IBD and ITP is rare, but it offers a therapeutic advantage as medications effective against both conditions can be used simultaneously to benefit the patient. Given the rarity of this association, we aim to describe a young patient of UC who presented with ITP as a rare extra-intestinal manifestation.

CASE REPORT

A 21-year-old male, working as a local driver and residing in a rural area of Kashmir, India, presented with a one-month history of fever and three days of loose stools, accompanied by hematochezia. The fever was low-grade, continuous, and associated with sweating. The stools were consistently loose, suggestive of large bowel involvement, mixed with fresh blood. They were associated with diffuse abdominal pain and tenesmus. A short course of intravenous antibiotics was prescribed to him at a peripheral health center before reporting to our hospital, but they did not relieve his symptoms much. The patient had a significant history of intermittent persistent diarrhea, bleeding per rectum, and hematuria on-off for the last 4 years. He was evaluated for the same on an outpatient department basis previously. Labs showed thrombocytopenia with giant platelets on PBF (peripheral blood film). PR proctoscopy revealed multiple small erythematous ulcers with friability, a biopsy showing partially ulcerated rectal mucosa with underlying normal mucosal glands and lamina propria revealing dense, chronic inflammatory cell infiltrate forming lymphoid follicles along with edema and congestion. He had previously received symptomatic treatment without a definitive diagnosis. During this admission to the ER, the patient was dehydrated and febrile to touch. He had tachycardia (110/min, regular) with BP of 100/70 mmHg and S02 of 97% on ambient air. The abdomen had diffuse tenderness on deep palpation without any definite signs of peritonitis. Labs showed thrombocytopenia, high CRP/ESR, and mild transaminases (Table 1).

PBF showed giant platelets with a manual platelet count of 60,000 per microliter. Normal gut flora was isolated from stool culture. ANA by Hep-2 cell lines was negative. Triple serology was negative. Ultrasonography of the abdomen showed mild splenomegaly (12 cm), and the rest was unremarkable. An initial impression of acute infectious enterocolitis (vs a flare of IBD) was made, and the patient was started on broad-spectrum intravenous antibiotics using ceftriaxone 1 g twice a day and metronidazole 500 mg thrice a day. The patient's clinical status, as assessed by the Truelove-Wittz severity index

Table 1. Laboratory parameters

Variables	Nov 2019	Feb 2020	Aug 06, 2023 (Admission)	Aug 11, 2023 (Discharge)
Hemoglobin (g/dL)	14.9	14.2	13.0	13.4
Platelets (per microliter)	22,000	50,000	34,000	60,000 - 100,000
ESR (mm/hour)	65	50	45	20
C-reactive protein	+	-	+	-
AST/ALT (IU/L)	N/A	N/A	82/52	42/20

ESR: erythrocyte sedimentation rate, AST: aspartate aminotransferase, ALT: alanine aminotransferase.

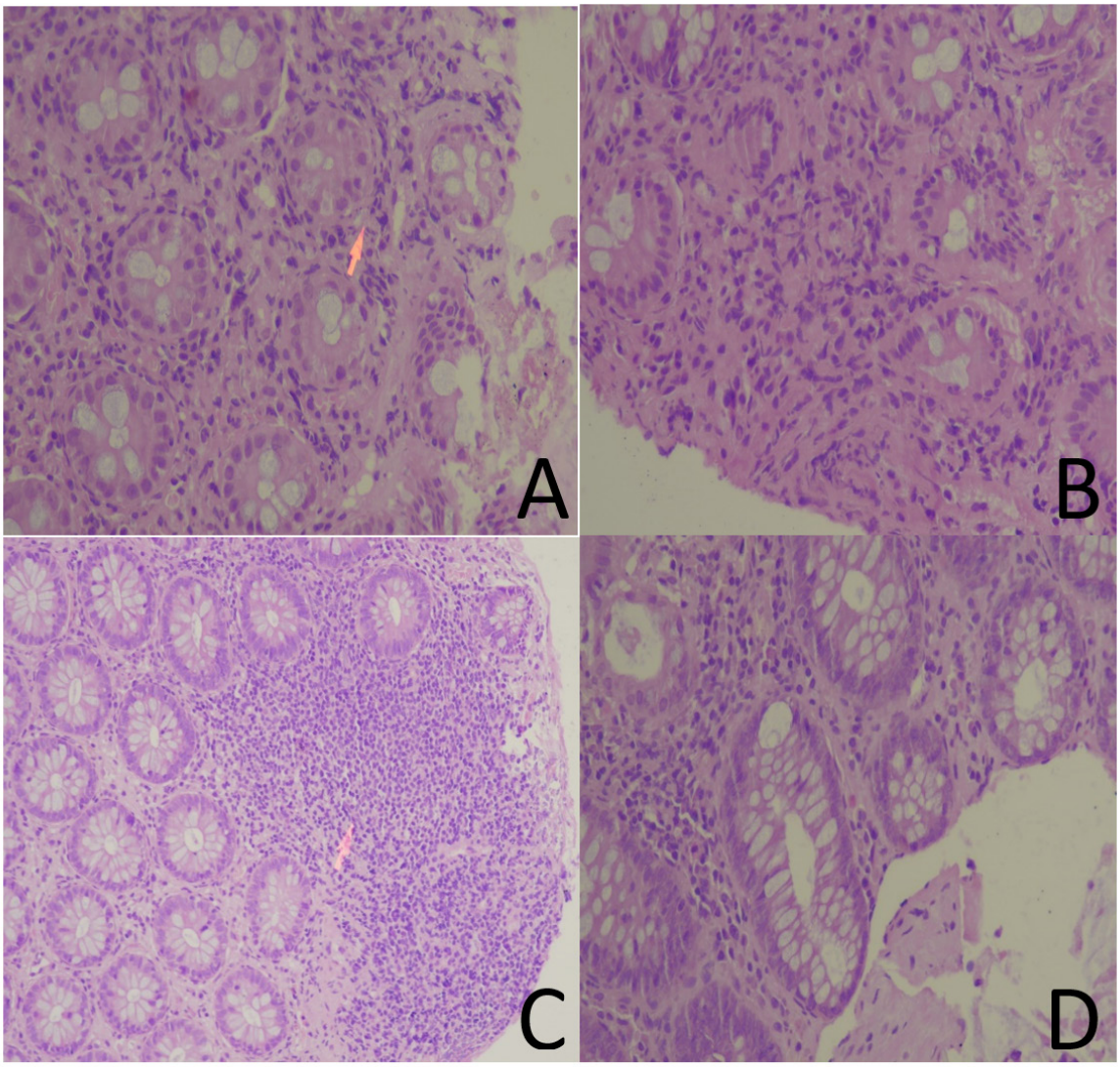

Figure 1. Colonoscopic biopsy showing A: cryptitis; B: crypt atrophy mucodepletion; C: lymphoid aggregate; D: basal plasma cell gradient

for UC, improved from severe to moderate during hospitalization. However, considering the patient's persistent symptoms along with a significant history, he was subjected to a colonoscopy. There was evidence of multiple small erosions and aphthous ulcers with background erythema from the caecum to the sigmoid colon. Severity was higher on the left side with normal vascularity and absent granularity, consistent with a Mayo endoscopic score of 3. Based on colonoscopy findings, an initial diagnosis of pancolitis with normal ileum and rectal sparing was made, and biopsies were taken for histopathological examination. The latter revealed edematous and congested lamina propria infiltrated by mixed inflammatory infiltrate and predominantly chronic mononuclear eosinophils with occasional cryptitis and crypt distortion foci. Additional pathological findings were found regarding basal plasma cell gradient, focal muco-depletion, and Paneth cell metaplasia. These findings, including focal mucodepletion and Paneth cell metaplasia, suggest chronic inflammation and disease activity in UC (Figure 1). A final impression of chronic UC with mild activity was made on this biopsy report. The same was reviewed with a gastroenterologist who started the patient on tapering doses of oral steroids and oral mesalamine 1.2 g twice a day. A hematology consultation was made because of long-standing thrombocytopenia. However, considering the patient's whole presentation, bone marrow findings ruled out cytotoxic causes of thrombocytopenia, supporting a diagnosis of peripheral platelet destruction consistent with ITP. Table 1 summarizes the laboratory findings upon admission.

The patient was discharged in a stable condition

after five days of hospitalization, with a platelet cunt of 100,000/µL. He continues to be monitored by gastroenterology and haematology specialists to ensure sustained remission and prevent complications.

DISCUSSION

As previously discussed, IBD is associated with numerous extra-intestinal manifestations. Patients with IBD also face an increased risk of developing other autoimmune diseases as well due to their overlapping pathogenic mechanisms.[7,8] ITP is considered a rare extra-intestinal manifestation of IBD.[9] Approximately 40 cases of this rare association have been documented in the literature. In such cases, treatment options include biologicals such as rituximab and infliximab and immunosuppressants like azathioprine.[10] Azathioprine is commonly employed as maintenance therapy in IBD. However, its use is associated with significant adverse effects such as myelosuppression, hepatotoxicity, and pancreatitis. These biologics target specific immune pathways, such as TNF-α inhibition (infliximab) or B-cell depletion (rituximab), to control IBD and ITP.

The coexistence of ITP as an extra-intestinal manifestation of IBD is exceedingly rare, making its recognition crucial for timely diagnosis and treatment. This case highlights the rare association of ITP as an extra-intestinal manifestation of IBD, emphasizing the need for clinicians to maintain a high index of suspicion of this association. Further studies and case reports are needed to elucidate the underlying mechanisms and optimize treatment strategies for patients with IBD and ITP.

CONCLUSIONS

ITP is a rare extra-intestinal manifestation in patients with UC. In a relevant clinical setting, thrombocytopenia should prompt physicians to consider this rare association, enabling timely diagnosis of both conditions and initiating treatments that can address both diseases concurrently.

Conflict of Interest

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Ethical Statement

No Approval is required letter from the Institutional Ethical Committee of Government Medical College Srinagar, Kashmir, India vide Ref. No. IRBGMC-SGR/MED/860dated 21/11/2024

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Authors' Contribution

Study Conception: MM, AR, BK, SB; Study Design: MM, AR, BK, SB; Literature Review: MM, AR, BK, SB; Critical Review: MM, AR, BK, SB; Manuscript preparing: MM, AR, BK, SB;

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