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A Case of Cerebral Venous Sinus Thrombosis Presenting During Relapse of Ulcerative Colitis

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Conflict of interest: None declared

Patient: Male, 27
Final Diagnosis: Cerebral venous sinus thrombosis
Symptoms: Headache • seizure • weakness
Medication: —
Clinical Procedure: —
Specialty: Gastroenterology and Hepatology

Objective: Rare disease

Background: Extra-intestinal manifestations of inflammatory bowel disease (IBD) include thromboembolic events that can present as deep vein thrombosis, pulmonary embolism, and cerebral venous sinus thrombosis. Cerebral venous sinus thrombosis is a rare complication of IBD that can be associated with high morbidity and mortality. This report is of a case of cerebral venous sinus thrombosis presenting in a young man during a relapse of ulcerative colitis (UC).

Case Report: A 27-year-old man presented with seizures and focal neurological deficit during a relapse of chronic UC. He was found to have left cerebral venous sinus thrombosis complicated by left frontotemporal infarction that was treated with anticoagulation therapy.

Conclusions: Thromboembolic events are well documented extra-intestinal manifestation of IBD. Cerebral venous sinus thrombosis is a rare but serious complication that can be fatal. The correct diagnosis and timely management require a high degree of suspicion in patients with IBD who present with a new-onset headache, focal neurological symptoms, seizure, or altered mental status.

MeSH Keywords: Cerebral Veins • Colitis, Ulcerative • Venous Thrombosis

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Background

Inflammatory bowel disease (IBD) is believed to have an autoimmune cause and can result in inflammation of the entire gastrointestinal tract, in Crohn's disease, or may mainly affect the colon, in ulcerative colitis (UC). Patients with IBD can present with diarrhea and rectal bleeding. Management of IBD involves treatment with 5-aminosalicylic acid (5-ASA), corticosteroids, and immunosuppression therapy.

IBD can be associated with both intestinal and extra-intestinal manifestations. Patients with IBD are at increased risk for thromboembolic events [1–4]. The risk of venous thromboembolism has been estimated to be raised by up to threefold when compared with the general population, even after the correction of abnormalities of pro-thrombotic factors [5,6]. Deep vein thrombosis (DVT) and pulmonary embolism (PE) are the most common forms of thromboembolic events associated with IBD [3,7]. Cerebral venous sinus thrombosis is a well-documented cause of acute onset neurological dysfunction in patients with IBD. Cerebral venous sinus thrombosis has been reported in patients with IBD during acute relapse [8].

Case Report

A 27-year-old man with a history of ulcerative colitis (UC), but no prior history of cerebrovascular accident (CVA) or transient ischemic attack (TIA), who was previously treated with 5-aminosalicylic acid (mesalamine), and 6-mercaptopurine presented to the emergency room with acute onset of right-sided weakness. He had recently been admitted to hospital for relapse of UC, which was treated with a ten-day course of steroid therapy. He continued to have bloody bowel movements after he was discharged from the hospital. He then developed a gradual onset of right-sided numbness and weakness that was associated with episodes of generalized seizures and persistent headache.

On physical examination, the patient was found to have reduced strength (2/5), diminished sensation, and brisk reflexes on the right side. Computed tomography (CT) and magnetic resonance imaging (MRI) of the head and brain showed a left paramedian frontoparietal cortical infarct secondary to cerebral venous sinus thrombosis (Figures 1, 2). A magnetic resonance venogram (MRV) of the head showed thrombosis of the left posterior frontal and anterior parietal cerebral cortex veins near the vertex with associated interruption of venous flow (Figure 3). There was no evidence of deep vein thrombosis of the lower extremities on Doppler ultrasound. A transthoracic contrast-enhanced echocardiogram showed a normal ejection fraction with no intracardiac thrombus or patency of the foramen ovale.

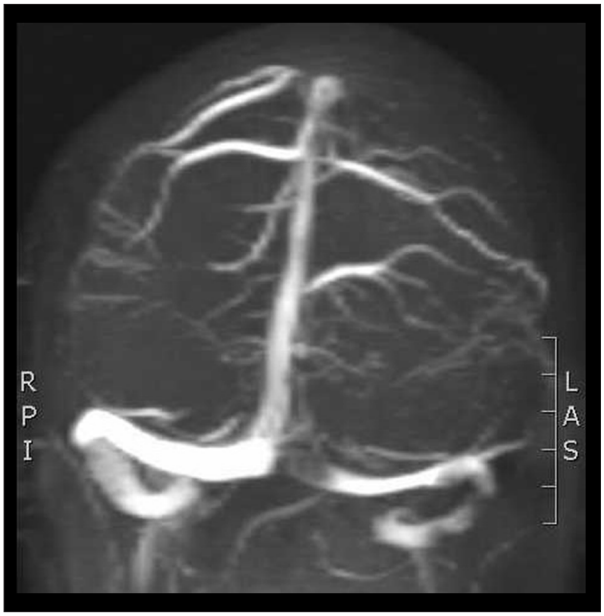

Figure 1. Infarction of the parafalcine frontoparietal lobe on brain magnetic resonance imaging (MRI) with fluid attenuation inversion recovery (FLAIR).

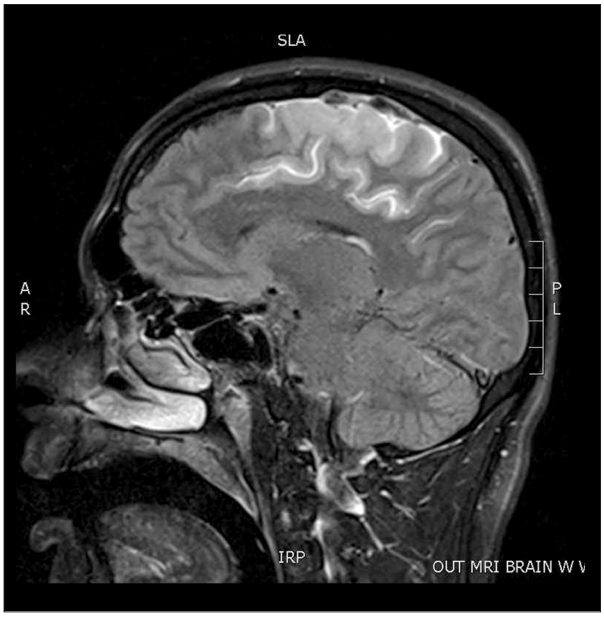

Figure 2. Infarction of the parafalcine frontoparietal lobe on brain magnetic resonance imaging (MRI) diffusion-weighted imaging (DWI).

Laboratory investigations showed normal renal function and glycated hemoglobin (HbA1c) of 6.5%. Serum lipid profiles and liver function tests were within normal limits. A limited coagulopathy workup was performed and was significant for a slightly increased international normalized ratio (INR) of 1.2, but was otherwise unremarkable. Laboratory investigations included investigations for factor V Leiden mutation, complement

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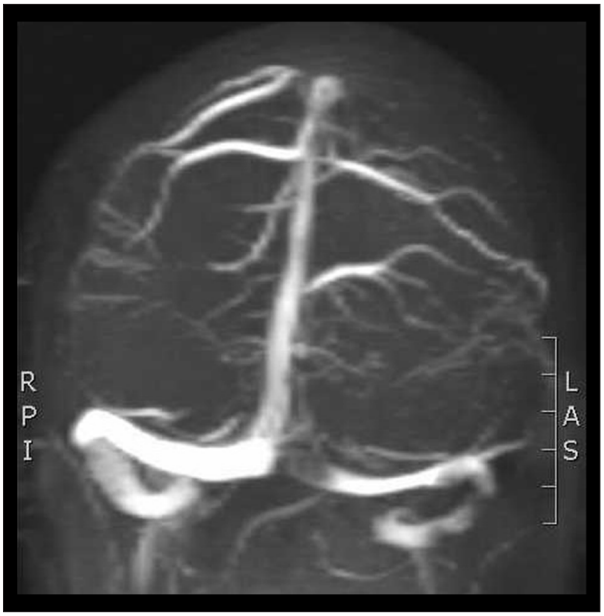

Figure 3. Interruption of venous flow on the left side of the sagittal sinus on magnetic resonance venography (MRV).

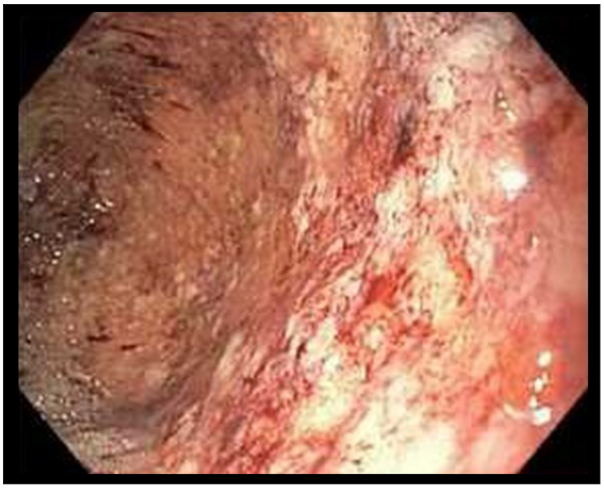

Figure 4. Severe mucosal ulceration of the distal colon, consistent with ulcerative colitis (UC).

factors, proteins C and S levels. Diffuse shallow ulcerations were noted on bedside flexible sigmoidoscopy, consistent with severe UC with a Mayo score or Disease Activity Index (DAI) of 11 (Figure 4).

The patient was managed with steroid therapy, 5-aminosalicylic acid, and infliximab. Treatment commenced with heparin infusion and the anti-epileptic, levetiracetam to manage the cerebral venous sinus thrombosis and seizures, respectively. His condition gradually improved and he had a reduced number of bloody bowel movements. He was discharged from hospital to a rehabilitation facility and was prescribed the anticoagulant, apixaban. He continued with multi-disciplinary management with follow-up at a gastroenterology and neurology outpatient clinics.

Discussion

The association between inflammatory bowel disease (IBD) and venous thromboembolism was first described by Bargen et al. in 1936 [9]. Aseptic cerebral venous sinus thrombosis is a rare and serious thromboembolic complication of IBD [3]. In IBD, cerebral venous sinus thrombosis is seen more commonly in male patients and at a younger age when compared with cerebral venous thrombosis secondary to other etiologies, and sagittal and lateral venous sinuses are more affected in patients with IBD [7,10]. The underlying pathophysiological process is not well established but is thought to involve altered coagulation cascade and a hypercoagulable state due to inflammation [11–13]. Hypercoagulable states, including hyperhomocysteinemia and mutation of factor V Leiden, have been reported in patients with IBD, but no direct correlation with cerebral venous sinus thrombosis has previously been reported [14,15]

The clinical picture of cerebral venous sinus thrombosis can be variable, but patients classically present with a new-onset headache [16,17]. Other reported symptoms include seizures and focal neurological deficits in the form of monoparesis or hemiparesis, in addition to features of increased intracranial pressure, confusion, and altered mental state [16,18–20]. A high degree of clinical suspicion is needed in patients with IBD who present with new-onset neurological symptoms to prevent delay in the diagnosis of cerebral venous sinus thrombosis. Screening and evaluation for prothrombotic conditions are recommended as part of the initial clinical workup [21,22].

Imaging studies can confirm the diagnosis of cerebral venous sinus thrombosis. Head computed tomography (CT) without contrast lacks sensitivity, but can identify associated hemorrhagic infarction that may exclude the use of anticoagulation therapy. Intracerebral hemorrhage can present in up to 30% of cases and is associated with poor clinical outcomes [20,23,24]. CT venography allows for rapid and reliable confirmation of cerebral venous sinus thrombosis [25,26]. Magnetic resonance imaging (MRI) with and without contrast and magnetic resonance venography (MRV) are the gold standard for diagnosis of cerebral venous sinus thrombosis that allow for direct visualization of the thrombus [22,27,28].

Patients with a confirmed diagnosis of cerebral venous sinus thrombosis should be treated with short-term anticoagulation therapy for three to six months, in the absence of contraindications [16,29,30]. Recurrence of cerebral venous sinus thrombosis or other forms of thromboembolic events is generally low, and long-term therapy should only be considered

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in a selected group of patients [31]. There is insufficient evidence to support the use of thrombolytic therapy in patients with cerebral venous sinus thrombosis, due to the lack of large randomized controlled trials [32,33].

Conclusions

Thromboembolic events are a well-documented extra-intestinal manifestation of inflammatory bowel disease (IBD). Cerebral venous sinus thrombosis is a rare event that can be life-threatening. As this case report has demonstrated, a high degree of clinical suspicion is needed to facilitate early diagnosis and prompt treatment.

Conflict of interest

None.

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