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## Case Report

# Successful management of dural venous sinus thrombosis secondary to ulcerative colitis in a pediatric patient: A case report ☆☆☆

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### ARTICLE INFO

#### Article history:

Received 4 March 2022

Revised 11 March 2022

Accepted 12 March 2022

#### Keywords:

Sinus thrombosis

Dural venous sinus thrombosis

Ulcerative colitis

Inflammatory bowel disease

Hypercoagulable state

### ABSTRACT

Cerebral venous sinus thrombosis secondary to inflammatory bowel disease is a clinically rare and challenging entity with serious sequela. We present a case of a 15-year-old female patient who was recently diagnosed with ulcerative colitis and had been suffering from headache for 4 days duration. During the diagnostic workup, computed tomography (CT) venography revealed Dural venous sinus thrombosis in the left transverse sinus extending into the left sigmoid sinus and the upper third of the left internal jugular vein as well as into the sinus confluence with non-occlusive filling defects in the superior sagittal sinus. Anticoagulant therapy with enoxaparin was initiated and the patient is being monitored in an outpatient setting regularly. Post-discharge disease course was uneventful. CT venography performed after 3 months illustrated partial recanalization of both left transverse and sigmoid sinuses. CVST is a rare extraintestinal manifestation of ulcerative colitis with significant morbidity and mortality which requires a high level of suspicion to establish a clear diagnosis. In spite of the fact that CVST is rare, it should be ruled out in inflammatory bowel disease patients with new onset seizures, headache, along with focal, and non-focal neurologic symptoms.

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☆ Acknowledgments: None.

☆☆ Competing Interests: The authors declare no conflict of interest. The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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<https://doi.org/10.1016/j.radcr.2022.03.049>

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## Introduction

Inflammatory bowel diseases are chronic inflammatory idiopathic disorders of the gastrointestinal tract, affecting almost 1.5 million people in north America, and 2 million in Europe [1,2]. The worldwide prevalence is exceeding 0.3%, with a rising trend in newly industrialized communities [3]. It has a significant impact on individual's quality of life posing mental, physical, and economic burden [4]. IBD expenditures are strikingly high amounting up to 5.6 billion euros every year in Europe alone [1]. Despite the differences between ulcerative colitis and Crohn's disease, both have a risk of venous thromboembolism (VTE); a serious extraintestinal, manifestation that has been linked to a worse prognosis [5]. In comparison to the general population, IBD patients have a 2- to 3-fold increased incidence of VTE [6]. Multiple hypothesized pathways were proposed to explain the pathophysiology of VTE episodes in IBD [7]. Fibrinolysis impairment, platelet activation, and endothelial dysfunction may all play a role [8]. Cerebral vasculature involvement in IBD is a rare consequence but rather a serious one, with few reports in the literature.

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## Case presentation

A 15-year-old Mediterranean female patient presented to the pediatric emergency unit at our center with a complaint of headache for 4 days duration. It started suddenly in the left frontal area which was non-progressive and radiating to the ipsilateral eye. The headache was associated with photophobia, nausea, vomiting, chills, hypoactivity, sleepiness, in addition to back, shoulder, and neck pain. The patient was recently diagnosed with ulcerative colitis (proctitis) and she is currently on mesalamine, omeprazole, and iron supplements. One week earlier to this event, she complained of bloody diarrhea, vomiting, symptomatic anemia, and significant weight loss in the last 2 months mainly reassembling an active flare of ulcerative colitis which was treated accordingly. She was transfused with 1 unit of red blood cells as her hemoglobin level was 8 mg/dL. Multiple episodes of vomiting were reported following admission that was relieved with ondansetron, and the headache was responsive to paracetamol. Complete blood count and basic metabolic profile only showed mild anemia (Hemoglobin: 10.0 g/dL, Hematocrit: 29.7%, Mean cell volume: 88.2  $\mu\text{m}^3$ ). Brain computed tomography (CT) scan with intravenous contrast showed that both cerebral hemispheres are within normal limits with normal gray-white matter differentiation. The brainstem and cerebellum are within normal limits, and the ventricles are bilaterally symmetrical. There is no mass or mass effect, intra-or extra-axial fluid collections or intracranial hemorrhage. The calvarium is intact with no fracture lines seen. CT venography showed filling defects observed in the upper third of the left internal jugular vein, the left transverse sinus, and extending to the left sigmoid sinus with sinus confluence involvement (Figs. 1A and 1B). There are also non-occlusive filling defects seen in the superior sagittal sinus. Other sinuses were opacified with no filling defects seen. All these findings are consistent with

Dural venous sinus thrombosis. Consequently, anticoagulant therapy; enoxaparin (4 mg, Q12) was started. CT venography performed after 3 months illustrated partial recanalization of both left transverse and sigmoid sinuses (Fig. 1C).

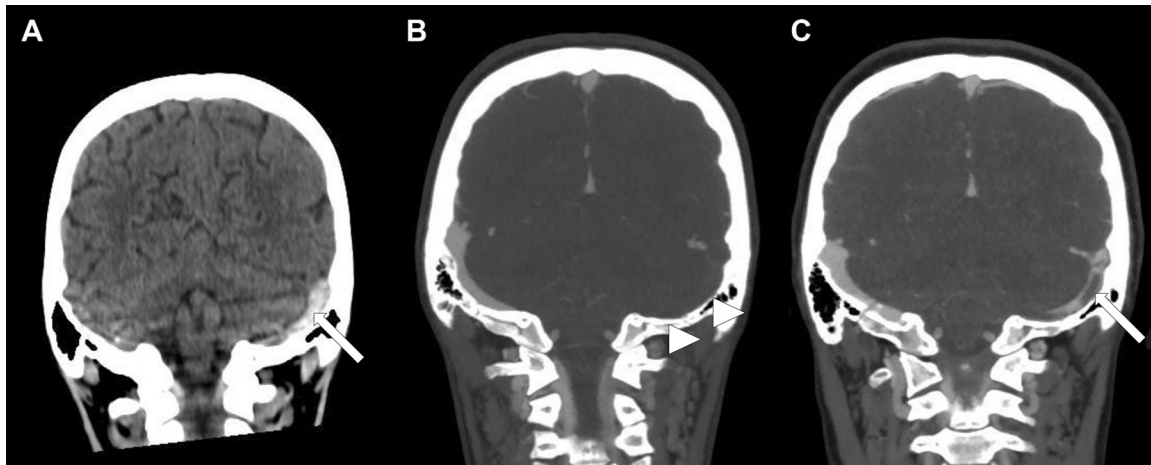
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## Discussion

Despite the fact that crosstalk between inflammation and thrombosis is well-known, deep evaluation of such interconnection has been only popularized in recent years [9,10]. Overwhelming evidence illustrated the pathophysiological underpinnings of inflammation-induced thrombosis in a multifactorial system incorporating proinflammatory chemokines, cytokines, endothelium, platelets, tissue factor, and even microparticles [10]. The mainstream hypothesis concludes that endothelial damage is the core agent in such process reassembled by the loss of vasodilatory and anticoagulant properties of vascular endothelial lining. Unfortunately, hypercoagulable state in inflammatory environments can develop without endothelial involvement, doubting the proposed mechanism [10]. Not surprisingly, thromboembolic accidents are one of the most common causes of, mortality and morbidity, in inflammatory diseases [11]. Many systemic inflammatory disorders exhibit hypercoagulability including systemic lupus erythematosus [12], familial Mediterranean fever [13], rheumatoid arthritis [14], vasculitis spectrum [15], and IBD [16]. The latter being the paradigm disorder showing how a confined inflammatory disorder increases thrombosis tendency in acute and chronic time frames [11].

In 1936, Bagen et al. published the first observation about the linkage between IBD, and VTE among 18 patients seen in Mayo Clinic out of 1000 IBD patients [17]. Following that time, a growing body of literature showed that IBD patients are more prone to thromboembolic accidents in its arterial and venous forms [18]. The prevalence of such events is about 1%-8% in clinical settings but necroscopy scans reported a prevalence of more than 40% [16,19]. The exact mechanism is poorly understood. IBD related arterial thromboembolism occurs mainly in the cerebral artery, aorta, mesenteric artery and retinal artery compared to the deep venous system thrombosis as in the lower extremities and lungs [20–24]. Although, these events remain a serious and life threatening extraintestinal manifestations of IBD, poor prophylactic measures are still reported [25]. A 10-year retrospective study including 1253 IBD patients revealed a significant lower mean age of IBD patients with thromboembolism (age: 53) compared to non-IBD cohort (age: 64) [26]. Further evaluation of IBD patients with thromboembolism suggest a male predominance with some controversy regarding the effect of pregnancy [23,27–29]. Active, extensive, and complicated IBD course is correlated with higher rates of developing thrombosis [16]. In contrast, almost one-third of IBD patients with thromboembolic events have quiescent disease [30]. The overall mortality is about 25% per episode, implying the need for robust prophylactic, and interventional acts [31].

As a well-recognized entity yet an underestimated one; pediatrics related cerebral, venous sinus, thrombosis, (CVST) remains a clinically challenging disorder. Its incidence is



**Fig. 1 – Coronal non-enhanced CT scan showing hyperdense left transverse and sigmoid sinus marked by the arrow (A). CT venogram coronal view demonstrating absence of contrast filling in the left transverse and sigmoid sinuses (arrow heads); consistent with venous sinus thrombosis (B). CT venogram coronal view after 3 months follow-up demonstrating partial recanalization of the left transverse and sigmoid sinuses as marked by the arrow (C).**

approximately 2.6 and 0.4-0.7 per 100,000 individuals per year in neonates and pediatric age groups respectively [32]. In these populations, CVST has a subtle presentation and requires a high level of suspicion. Clinical presentation varies which could include seizures, encephalopathy, headache, nausea, vomiting and other focal and non-focal neurologic manifestations [32]. Sinus thrombosis is diagnosed by a group of imaging modalities. Associated hemorrhagic complications should be excluded utilizing CT scan without contrast despite its poor sensitivity. MR venography is the diagnostic gold standard imaging for direct precise visualization of venous thrombosis while CT venography presents a robust method for fast diagnosis. Magnetic resonance imaging with contrast and cerebral detailed 3-dimensional reconstruction is readily available especially when large volume of tissues is scanned during vascular peak enhancement [33–35]. Therapeutic plan of CVST should be articulated and implemented in a fast manner. Firstly, underlying predisposing disorder should be controlled if known. Neurologic complications as in intracranial hypertension and seizure should be managed followed by the administering of antithrombotic and anticoagulant agents preventing the propagation of the thrombus. Prophylactic measures and good disease control aids in preventing future attacks as previously described [35].

Although IBD related CVST is well illustrated in adults, reports regarding the pediatric age group are scant. Martín-Masot et al. reported a transverse sinus thrombosis in a 5-year-old boy with ulcerative colitis following colectomy hence highlighting the importance of thrombosis prophylaxis especially in the postoperative period [36]. Ashrafi et al. reported an 11-year-old male who was recently diagnosed with ulcerative colitis and presented with pseudotumor cerebri secondary to superior sagittal sinus thrombosis during a flare [37]. Moreover, Liu et al. described a 12-year-old child with newly diagnosed ulcerative colitis who presented with sudden left-sided hemiparesis and found to have cerebral sinovenous thrombosis [38]. In addition, Robert et al. demonstrated a case of

a 14-year-old boy with CVST, and ulcerative colitis diagnosed by MRI [39]. Sometimes superimposed genetic hypercoagulable disorders can aggravate IBD related vascular events as in a 10-year-old boy with ulcerative colitis and CVST that was found to have heterozygous factor V Leiden [40]. Multiple sinus involvements were also reported as in a 10-year-old girl who presented with headache and seizures and later diagnosed with superior sagittal, right transverse, sigmoid sinus thrombosis as a sequelae of ulcerative colitis [41].

## Conclusion

CVST is a rare extraintestinal manifestation of IBD with significant morbidity and mortality rates which requires a high level of suspicion to establish a clear diagnosis. In spite the fact that CVST is rare, it should be ruled out in IBD pediatric patients with new onset seizures, headache, along with focal, and non-focal neurologic symptoms.

## Funding

There is no funding to report.

## Availability of data and materials

Not applicable.

## Ethics approval

This report has been conducted and written in accordance to the ongoing regulations for case reports and case series in the

king Abdullah university hospital (KAUH). IRB approval (Ref: 79/147/2022) waived the need for written informed consent with total anonymization of the patient's identity.

## Patient consent

Written informed consent was obtained from the patient for publication of this report and any associated images.

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