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Ulcerative colitis and cerebral venous thrombosis: A case report with literature review

Nesrine Kouki[1]*; Hanene Ben Rhouma[1,2]; Aida Rouissi[1,2]; Thouraya Ben Younes[1,2]; Hedia Klaa[1,2]; Ichraf Kraoua[1,2]; Ilhem Ben Youssef-Turki [1,2]

[1]Department of Child and Adolescent Neurology, Research laboratory LR18SP04, National Institute Mongi Ben Hmida of Neurology, Tunis, Tunisia.

[2]University of Tunis El Manar, Faculty of Medicine of Tunis, Tunis 1007, Tunisia.

***Corresponding Author: Nesrine Kouki**
Department of Child and Adolescent Neurology, Research laboratory LR18SP04, National Institute Mongi Ben Hmida of Neurology, Tunis, Tunisia.
Tel: +216 50 563 180;
Email: nesrine_kouki@yahoo.com

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Abstract

Background: Cerebral Venous Thrombosis is a rare complication of Inflammatory Bowel Disease and the pathogenesis underlying its incidence remains enigmatic.

Methods: We present a case of a child with a history of ulcerative colitis complicated with Cerebral venous thrombosis, and through the search engine 'Pubmed' we performed literature review to select similar cases in Pediatric population.

Results: In our case, Cerebral Venous Thrombosis was revealed by intense sharp headache just few days after a severe relapse under high oral doses of steroids and sulfasalazine. A cerebral Magnetic Resonance Imaging and Magnetic Resonance Angiography (MRA) showed an extended thrombosis complicated with cerebral infarction and hemorrhage.

The literature review showed that most Cerebral Venous Thrombosis came during or few months after a severe relapse incriminating a prothrombotic state secondary to systemic inflammation. Undergoing drugs intake was absent almost in half cases. Radiological findings supported the association with enlarged cerebral thrombosis.

Conclusion: Inflammatory Bowel Disease patients are associated with higher risk of thromboembolic complications. Early recognition and onset of the appropriate treatment prevent progression and complications which may be lethal.

Keywords: cerebral venous thrombosis; ulcerative colitis; headache; children.

Abbreviations: CVT: Cerebral Venous Thrombosis; IBD: Inflammatory Bowel Disorders; MRA: Magnetic Resonance Angiography; NR: Normal Range; NM: Not Mentioned; SSS: Superior Sagittal Sinus; UC: Ulcerative Colitis; VTE: Venous Thromboembolism; INR: International Normalized Ratio.

Introduction

Cerebral venous thrombosis is a rare but a life threatening complication of inflammatory Bowel disease notably in ulcerative colitis.

Patients could suffer of this complication during or after a relapse of the disease, either under adjusted treatment or without any medications, the pathogenesis underlying its incidence remains enigmatic, therefore primary prevention is a laborious task for the physicians. Clinical manifestations of CVT are variable and include headaches, vomiting, focal neurologic deficit, and decreased level of consciousness. Here, we describe the atypical clinical presentation and treatment of a young man with CVT associated with UC.

Case report

A 16 year-old teenager born from a non-consanguineous marriage had a family history of type 1 diabetes in the sister. He had no significant antenatal or perinatal history. Psychomotor development was normal. He had no particular health complaints until 2020. At the age of 15, he started suffering from prolonged bloody stools. After biological and endoscopic explorations and based on the findings of intestinal tissue biopsy, the diagnosis of ulcerative colitis was made on January 2021 and the patient was put under an anti inflammatory combination of oral and rectal suppository treatment. The clinical remission was short and the patient was admitted in gastro-intestinal department on February 2021 for a severe relapse. The evolution was favorable under intravenous corticosteroids intake. The patient was discharged 3 weeks later with a medical prescription of oral high doses of steroids and oral sulfasalazine.

One week later, he was referred to the neurological emergency department for acute severe and sharp headache that started 3 days ago. Headache was constructive, localized on the left hemicranial side, and preventing the sleep, with no respond to paracetamol intake. He had no blurred vision or vomiting.

The neurological examination showed no abnormalities. A cerebral Magnetic Resonance Imaging (MRI) and Magnetic Resonance Angiography (MRA) were performed and showed an extended thrombosis of the right sigmoid and transverse sinuses reaching the internal jugular vein complicated with a cerebral infarction of the right temporoparietal lobes with hemorrhage (Figure 1).

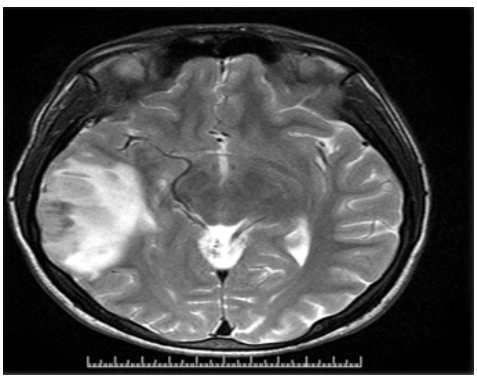

Figure 1: Axial T2 weighted MRI sequencing showing intracerebral hemorrhage in the right temporoparietal lobes.

Laboratory findings showed: An elevated white blood cells count at 20,000/mm^3 (Normal Range (NR): 4.000-10.000/mm^3), a hypochromic Anemia at 7 g/dl (NR: 12.5-17.5 g/dl), a normal platelets rate, CRP was normal, liver and renal function were within normal limits.

He was admitted in pediatric neurological department. The patient had prothrombotic assessment for antithrombin III, protein S, protein C deficiencies and anti phospholipid antibody syndrome were normal, plasma homocysteine level was normal.

Anticoagulation treatment was initiated with intravenous unfractionated heparin infusion, and oral acenocoumarol was introduced 5 days after reaching an activated Partial thromboplastin time of 70 seconds.

During hospitalization, the evolution was favorable, the patient didn't develop any new symptoms and headache gradually improved.

He was discharged 2 Weeks later after a total recovery with an oral anticoagulation treatment based on acenocoumarol one tablet three times daily with an INR in the therapeutic range.

Discussion

Inflammatory Bowel Disease (IBD) has intestinal and extra intestinal manifestations which can affect central nervous system. In fact, IBD patients are at a 2 to 3 -fold higher risk of developing a Venous Thromboembolism (VTE) compared to the general population [1].

A precise incidence is not yet well described, but CVT seems to be more common in ulcerative colitis than in Crohn's disease [2].

The presence of headache or acute worsening of neurological status in a patient with IBD should alert clinicians about the possibility of CVT.

Only ten pediatric cases of CVT in ulcerative colitis patients were described in literature all summarized in Table 1.

In our case, cerebral venous thrombosis was revealed by intense isolated headache and consistent with the findings in the reviewed pediatric cases (Table 1). Indeed, based on the International classification of headache disorders 3rd edition, in CVT headache is by far the most frequent symptom and has no specific characteristics but most often, is diffuse and severe [3,4].

The pathogenesis of CVT relies on interrelated Factors. IBD is known to cause an effective systemic inflammation considered as a prothrombotic state [5] which is in its highest level during relapses explaining the short delay between relapse and clinical manifestation in the case we report. Aside from inflammation, other factors such as anemia, thrombophilia and corticoids intake are incriminated.

Screening for prothrombotic conditions is systematic considering its possible association with IBD and its contribution in determining the duration of oral anticoagulation.

Radiological findings revealed that the involvement of mul-

Table 1 : Ulcerative colitis and CVT in pediatric population reported in literature.

Investigations	Patient 1	Patient 2	Patient 3	Patient 4	Patient 5	Patient6	Patient 7	Patient 8	Patient 9	Patient 10	Our case
Age (years)	12	17	12	10	11	18	16	18	14	14	**16**
Gender	female	male	male	male	Female	male	female	male	female	male	**male**
Time between IBD diagnosis and CVT	Diagnosed with CVT	1.5 year	4 years	3 years	3 years	1 years	1.5 years	5.5 years	Diagnosed with CVT	Diagnosed with CVT	**3months**
Date of last relapse	Concomittant to CVT	3 weeks before	Concomittant to CVT	Concomittant to CVT	Concomittant to CVT	Concomittant to CVT	Concomittant to CVT	1 month before	Concomittant to CVT	Concomittant to CVT	**1 month before**
Severity of the last relapse	severe	moderate	NM	NM	NM	moderate	severe	severe	severe	severe	**severe**
Undergoing treatment	NM	No	NM	NM	NM	steroids intake	No	High doses of steroids +sulfasalazine	No	No	**High doses of steroids +sulfasalazine**
Clinical presentation	left-sided hemiparesis + numbness + intermittent convulsion	Acute Headache	Acute Headache	Acute Headache	Acute headache	Acute headache	Acute headache+ generalized tonic-clonic seizure	Acute headache	Paresthesia and partial motor deficit of the left lower limb	Headache+ hemiparesis +	**Acute headache**
Biological abnormalities	Hyperleukocytosis+moderate anemia	low protein S levels	NM	NM	NM	NM	mild thrombocytosis +anemia+high CRP	anemia	Hyperleukocytosis+ Hypertransaminasemia+high CRP	Hyperleukocytosis + anemia	**Hyperleukocytosis + anemia**
Immunological tests	negative	negative	negative	NM	NM	negative	negative	negative	negative	negative	**negative**
Radiological findings	VT of the SSS with secondary hemorrhage and infarction in frontal and temporal lobes	VT of the right transverse and sigmoid sinuses	NM	NM	NM	VT of the left lateral sinus	left lateral and sigmoid sinuses thrombosis with left temporal infarction	VT of the transverse sinus	VT of the sagittal sinus infarction	VT of the sigmoid and lateral sinuses extending to the internal jugular vein + hemorragic infarction	**extended thrombosis of the right sigmoid and transverse sinuses reaching the internal jugular vein complicated with a cerebral infarction of the right temporoparietal lobes with hemorrhage**
Evolution	complete recovery	complete recovery	complete recovery	Coma and Death	Death	complete recovery	complete recovery	complete recovery	complete recovery	complete recovery	**Complete recovery**
reference	Medecine , 2020 [10]	Brain & Development (2011) [11]	Journal of Digestive Diseases 2015 [12]			Revue de la medecine interne 2006[13]	Acta Gastro-enteroogica Belgica,2007[14]	World journal of Gastro-enterology ,2008[15]	Pediatrics, 2013 [16]	Journal of pediatric gastroenterologyand nutrition ,1989 [17]	

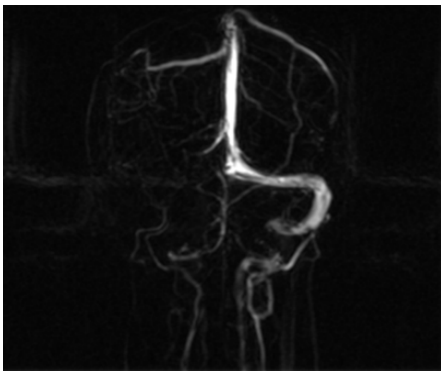

Figure 2: Venous MRA with contrast showing occlusion of the right transverse sinus and the internal jugular vein.

tiple sinuses and extensive venous clot were more associated with hemorrhagic infarction than those with nonhemorrhagic lesions (Table 1). Patients with haemorrhagic infarction usually have more disability after 3 months of evolution compared to those with non haemorrhagic lesions [6], luckily our patient only suffered from headache which improved rapidly after the onset of treatment.

The prothrombotic state in CVT is thought to explain the enlarged thrombosis [6], explaining the cerebral MRI results found in our patient with the clot reaching the internal jugular vein. (Figure 2).

In IBD, the underwent therapy interferes with the pathogenesis of CVT. Indeed, Corticosteroids as previously mentioned may induce a hypercoagulability state by activating the coagulation Factors FVII, VIII and XI and by blocking the anticoagulation mechanisms [7]. This adverse effect is described especially with high doses [8] which was the case of our patient. On the other hand, Aminosalicyclates such as Sulfasalazine may inhibit spontaneous and thrombin-induced platelet activation which has been thought to reduce VTE risks [9].

The current practice in the treatment of CVT in patient with IBD is similar to patients without IBD, but the duration may differ depending on the results of etiological investigation [7].

The screening of an associated prothrombotic disease was negative in our patient.

Our case represents an unusual complication of ulcerative colitis. Indeed, IBD is recognized as a risk factor for VTE. The underlying mechanisms are Interrelated but not well-elucidated. Acute onset of Neurological symptoms in IBD patients, should alarm clinicians to consider TVC diagnosis and start an early treatment minimizing complications that may be lethal.

Declarations

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Competing interests: The authors report no competing interests.

Ethics approval: Not applicable.

Informed consent: The patient authorized the writing and publication of this case report.

Author participations:

-Nesrine kouki; Hanene Ben Rhouma, Aida Rouissi, Thouraya Ben Younes, Hedia Klaa, Ichraf Kraoua and Ilhem Ben Youssef-Turki participated in taking care of the patient and the bibliography RESEARCH.

-Nesrine kouki and Hanene Ben Rhouma participated in the design and writing the final manuscript.

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