Clinical Case Report

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Ulcerative colitis with Guillain–Barré syndrome

A case report

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Abstract

Rationale: Ulcerative colitis is a chronic and recurrent inflammatory disease involving the intestine. It is reported that about 40% of patients with ulcerative colitis have extraintestinal manifestations, where as the literature on neurological involvement as extraintestinal manifestation is rather limited. Guillain-Barré syndrome is an abnormal immune-mediated and acute-acquired demyelinating disease that mainly affects the peripheral nervous system and often has a phenomenon of protein-cell separation of cerebrospinal fluid. Here, we report a rare case of ulcerative colitis with Guillain-Barré Syndrome.

Patient concerns: We described a patient with Guillain-Barré syndrome during the remission period of ulcerative colitis. Clinical manifestations are the numbness of the upper extremities, weakness in the limbs and the inability of the fingers companion. Cerebrospinal fluid (CSF) showed albuminocytological dissociation and electromyography suggested neurogenic lesion.

Diagnoses: Ulcerative colitis with Guillain-Barré syndrome was diagnosed based on the history of ulcerative colitis, related symptoms, typical cerebrospinal fluid albuminocytological dissociation and evidence of neurogenic injury through electromyography.

Interventions: The patient was treated with intravenous (IV) methylprednisolone.

Outcomes: After the treatment of glucocorticoid, the symptoms of the nervous system were disappeared.

Lessons: Neurological involvement of extraintestinal manifestation during the remission period of ulcerative colitis also exists in the clinic. This case highlights the need for diagnostic vigilance in cases of ulcerative colitis involving the peripheral nerves during the remission period. We recommend cerebrospinal fluid examination and electromyography in view of rare but serious possibility of Guillain-Barré syndrome.

Abbreviations: 5-ASA = 5-aminosalicylate, CSF = cerebrospinal fluid, EMG = electromyography, ESR = erythrocyte sedimentation rate, GBS = Guillain–Barré syndrome, TNF-α mAb = tumor necrosis factor-α monoclonal antibody, UC = ulcerative colitis.

Keywords: glucocorticoid, Guillain–Barré syndrome, ulcerative colitis

1. Introduction

The extraintestinal manifestations of ulcerative colitis (UC) mostly occur in the active phase, but the literature on neurological involvement as extraintestinal manifestation is rather limited. So far, there have been 4 case reports about Guillain–Barré syndrome (GBS) during a relapse of UC.[1–4] There are less case reports about UC with GBS during a remission of UC.

Here, we report a case of UC patient with GBS during a remission of UC in China, confirmed by cerebrospinal fluid (CSF) examination as well as electromyography. We believe this case report will ensure an accurate and prompt diagnosis of UC with GBS not only in the active period but also in remission of UC.

2. Case report

A 31-year-old female patient was admitted to our hospital for bloody and mucopurulent diarrhea for 3 months with body numbness and weakness for 10 days in 2017. Three months ago, the patient had no precipepating factors to occur bloody and mucopurulent diarrhea (3–5 times/day) with abdominal pain, but without tenesmus and fever. She went to the local hospital to undertake colonoscopy and it suggested severe UC, which was in active stage and involved the full colon. She was treated with 5-aminosalicylate (5-ASA) Mesalazine SR Granules (Etiasa) (oral administration [PO]) 250 mg 4 times per day, Mesalazine Slow Release Tablet (rectal administration) 1000 mg per night, Live Combined Bifidobacterium, and Enterococcus capsules (PO) 420 mg 3 times per day. She was discharged as soon as the improvement of clinical manifestations was achieved. Ten days ago, she was readmitted to the Department of Neurology of our hospital for numbness of upper extremities, weakness of limbs, and the symptoms aggravated and progressed from distal limbs to proximal limbs. The patient's hands began to show the inability of holding things, loss of abduction, and opposition of fingers gradually. At that time, she had no gastrointestinal symptoms such as bloody and mucopurulent diarrhea or abdominal pain. The patient denied other previous diseases such as hypertension, diabetes, hepatitis B, tuberculosis, and so on.

Editor: N/A.

Written informed consent was obtained from the patient for the publication of the case report.

The authors have no conflicts of interest to disclose.

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Table 1

The positive results of laboratory examinations in local hospital 3 months ago.

Laboratory work-up test	Results		Reference range
Blood routine	Hemoglobin (Hb)	106 g/L	110–150 g/L
Urine routine	Blood	3+	Negative
	Red blood cell	668.70/μL	0–28/μL
Stool routine	Occult blood (OB)	+	Negative
	Transferrin	+	Negative
D-dimer	D-dimer	0.56 mg/L	0–0.55 mg/L

Physical examination showed that vital signs were normal. On neurological examination, the patient was fully consciousness. No speech or articulation disorder was detected. The bilateral pupils were the same size and sensitive to the light reflect, eyes movement was normal, and eyes tremor was not observed. Bilateral nasolabial grooves were symmetrical. There was no tongue deviation during tongue protrusion. The muscle strength of bilateral upper limbs were at IV level (Lovett muscle grading standard) and symmetrical, and it was at IV level in bilateral proximal lower extremity and at III level in bilateral distal lower extremity. Superficial and deep sensory modalities of limbs were apparently normal. A moderate impairment of radioperiosteal reflex, knee reflex, and ankle reflex was observed. The patient could not complete the finger-to-nose test and Romberg test. Meningeal irritation sign and pyramidal signs were negative. Physical examination demonstrated a well-nourished patient with a normal abdomen.

Three months later, the patient exhibited positive results of laboratory examinations in local hospital, which are listed in the Table 1. Blood coagulation function test and biochemical profile were within normal range. Histologic examination showed rectal mucosal chronic and active inflammation with crypt abscess and cryptitis. Computed tomography (CT) scan of the whole abdomen showed that the spleen was slightly larger, there were stones in both kidneys, the duodenal descending diverticulum, and the rectum and colon were slightly thickened and strengthened, which may indicate inflammatory disease.

The patient's positive results of laboratory examinations in our hospital are listed in Table 2. Blood routine test, urine routine, stool routine, stool cultures, tumor markers, vitamin B_{12}, erythrocyte sedimentation rate (ESR), procalcitonin, anti-streptococcus hemolysin O antibody, rheumatoid factor (RF), and C-reactive protein were all in normal range. Electromyography (EMG) showed limb nerve conduction velocity was significantly slow, lower amplitude, F wave delay. Magnetic resonance imaging (MRI) of head and neck showed cervical degenerative changes; the right vertebral artery was thin. There were no obvious abnormalities in chest CT examination. Color ultrasound suggested that her spleen was larger than normal. CSF examination showed acid-fast staining, ink negative staining, and CSF culture were all negative.

UC with GBS was diagnosed on the basis of patient symptoms and typical CSF albuminocytological dissociation and electromyographic evidence of neurogenic injury. She accepted the treatment programs of neural protection, supplying folic acid and B12, and other supportive treatments. Concomitantly, she was treated with intravenous (IV) methylprednisolone 500 mg/day. After the treatment with methylprednisolone for 3 days, her limbs weakness was relieved, muscle strength was improved to V-level, and the feeling of numbness was also relieved. Five days later, the methylprednisolone was halved (250 mg/day, IV drip). And 8 days later, the methylprednisolone was halved again (125 mg/day, IV drip). After accepting the treatment of glucocorticoid for 11 days, methylprednisolone was displaced by Methylprednisolone tablets (Metsola) (60 mg, once daily, PO), and then muscle strength recovered completely to V level, the numbness of upper extremity disappeared completely, Methylprednisolone tablets was reduced by 10% every 2 weeks, and then she kept taking Methylprednisolone tablets 10 mg for 6 months. After the treatment with glucocorticoid for 11 days, the patient's condition was significantly improved. She was discharged and continued to take Methylprednisolone tablets orally at home.

3. Discussion

UC is a chronic and recurrent inflammatory disease involving the intestine.[5] About 40% of patients have extraintestinal manifestations,[6] such as recurrent oral aphthous ulcer, anterior uveitis, primary biliary cirrhosis, and ankylosing spondylitis, whereas there is less report about neurological involvement as extraintestinal manifestation. GBS is an abnormal immune-mediated and acute-acquired demyelinating disease that mainly affects the peripheral nerves and often has a phenomenon of albuminocytological dissociation of CSF. It is mostly a self-limited disease.[7] The pathogenesis of UC with GBS has not been elucidated, and there are several possible hypotheses.

As both UC and GBS are autoimmune diseases, there may be similar immunological mechanisms between these 2 diseases. Previously, GBS was considered as an extraintestinal manifesta-

Table 2

The positive results of laboratory examinations in our hospital.

Laboratory work-up test	Results		Reference range
Examination for anemia	Folic acid	↓3.29 ng/mL	>5.31 ng/mL
	Ferritin	↓6.3 ng/mL	10–291 ng/mL
ANA	ANA	1: 100	≤1:100
	ANA-mode	Granular (S)	
CSF cytology examination	White blood cell	1×10^6/L	$(0–8) \times 10^6$/L
	Red blood cell	0/L	0/L
	Ration of lymphocyte	0.68	0.7
	Ration of mononuclear cells	0.32	0.3
CSF biochemical examination	Glucose	2.60 mmol/L	2.5–4.5 mmol/L
	Protein	0.67 g/L	0.20–0.40 g/L
	Chlorine	98 mmol/L	120–130 mmol/L

CSF = cerebrospinal fluid.

tion of UC, which involves the nervous system.[8] The extraintestinal manifestations of UC often occur in the active period,[1–4] while GBS can occur in the remission of UC.[9] The exact pathogenesis of UC with GBS is unclear and it may be related to the following factors: UC-associated vasculitis,[6] postinfection immunity, malnutrition, toxic metabolites, vitamin deficiency, and thrombotic disease.[10]

However, Bouchra et al[6] do not consider GBS as a extraintestinal manifestation of UC and suggest that the occurrence of GBS is closely related to the use of biologic anti-tumor necrosis factor-α monoclonal antibody (anti-TNF-α mAb) in patients with UC. TNFα has an immunomodulatory function; it causes a decrease in inactivation of myelin-specific T cells and prolongs the survival time of activated T cells.[11] Anti-TNF-α mAb specifically binds to TNFα, breaking the balance between the TNFα receptor and the antibody in the peripheral nervous system, enhancing the myelin-specific T cell activity, and causing permanent immune damage to the peripheral nervous system; then, it causes the patients with UC who have immune dysfunction originally to have GBS.[12] The US Food and Drug Administration's Adverse Events Reporting System showed that 17 cases of GBS patients were associated with the use of anti-TNF-α mAb.[13] The British Rheumatism Association recommends that patients with a medical history of demyelination should avoid the use of anti-TNFα mAb.[14] However, there is a view that the pathogenesis of GBS is related to opportunistic infections such as the infection of respiratory virus and campylobacter, and the reason why the patients suffer from UC with GBS is that the TNFα mAb increases the risk of opportunistic infections in patients with UC.[12]

In this case, the patient was a young woman with GBS during the remission of UC, and her symptoms were relieved promptly after high-dose glucocorticoid treatment. This patient did not have a history of TNF-a treatment during UC onsets and did not have a previous history of viral infection. Therefore, GBS is thought to be an extraintestinal manifestation of UC that involves peripheral nervous system.

Author contributions

Resources: Shan Tian, Ke Zhou.
Supervision: Weiguo Dong.
Validation: Zhengru Liu, Shan Tian, Weiguo Dong.
Writing – original draft: Zhengru Liu, Shan Tian.
Writing – review & editing: Zhengru Liu.

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