CASE REPORT

An unusual presentation of ulcerative colitis

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SUMMARY

Ulcerative colitis (UC) may have diverse extraintestinal manifestations. Nutritional deficiencies, medications or immune-mediated epiphenomena are considered to be pathogenic mechanisms involved. We describe a case of a 56-year-old woman who presented with rapidly progressive tingling paraesthesias in both lower limbs followed by sensory ataxia, ascending spastic quadriparesis, urgency and urge incontinence of 3 months duration. She had an episode of bloody diarrhoea 1 month later for which a colonoscopy was carried out with colonic biopsy features revealing a diagnosis of UC. In view of lack of alternative aetiology, the posterolateral column disease in our patient was attributed to an extraintestinal manifestation of UC. She improved dramatically with a course of intravenous steroids followed by tapering dose of oral steroids and oral mesalamine for her UC. Although rare, UC should be considered in the differential diagnosis of posterolateral column disease of the spinal cord in the appropriate clinical scenario.

BACKGROUND

Although ulcerative colitis (UC) is primarily an inflammatory disease involving the colonic mucosa, it can also be considered a systemic disorder in view of its variable presentations. Extraintestinal manifestations of UC are relatively common with a lifetime risk of developing at least one of them being 50%.¹ Occasionally, UC may have prominent neurological manifestations, but UC presenting as a non-compressive posterolateral column disease is extremely rare. This case reinstates the golden dictum that extensive diagnostic evaluation is required to arrive at an aetiological diagnosis of non-compressive posterolateral column myelopathy in view of multiple differentials to be considered.

CASE PRESENTATION

A 56-year-old postmenopausal woman presented with rapidly progressive tingling paraesthesias in both lower limbs of 3 months duration. This was followed a month later by unsteadiness while walking (more at dark) and ascending weakness of hands and feet (distal>proximal) followed by autonomic symptoms in the form of urgency and urge incontinence. During the course of these symptoms she developed an episode of bloody diarrhoea lasting for a week, which subsided with a course of oral metronidazole therapy (400 mg three times a day for 7 days). No other systemic symptoms were observed. Physical examination revealed distal predominant weakness, spasticity, hyper-reflexia and bilateral extensor plantar response with pseudoathetosis. Posterior column dysfunction was noted in the

form of abnormal joint position response (distally), impaired vibration response up to acromion process bilaterally with positive Romberg sign as well. A syndromic diagnosis of non-compressive myelopathy (posterolateral column disease) was performed and the patient was subjected to a battery of investigations in order to yield the underlying aetiology.

INVESTIGATIONS

Baseline haematological parameters including peripheral blood film were normal except for an elevated erythrocyte sedimentation rate of 51 mm in the first hour (normal<30). Biochemical parameters (including blood glucose, renal function, liver function, sodium, potassium, calcium, magnesium, phosphorous and lipidogram) were within normal limits. Serum ACE levels were normal. Viral markers (HIV, human T cell lymphotropic virus 1, hepatitis B and hepatitis C virus) were negative. Autoimmune panel revealed positive antinuclear antibody in speckled pattern (3+). However, perinuclear antineutrophil cytoplasmic antibodies (p-ANCA), cytoplasmic ANCA (c-ANCA), anti-tissue transglutaminase, anti-ds DNA, anti-Ro and anti-La were negative. Serum vitamin B₁₂ levels were 1123 pg/mL (normal 320–1600 pg/mL). Serum and urine protein electrophoresis failed to reveal any abnormal monoclonal (M) band. Cerebrospinal fluid (CSF) analysis was acellular with elevated protein (155 mg/dL, normal 20–40 mg/dL) and normal sugar levels. CSF analysis for malignant cells and tuberculosis (TB-PCR) were negative. CSF oligoclonal band was positive on paired serum CSF sample analysis. Serum and CSF venereal disease research laboratory tests were both non-reactive. Gad-enhanced MRI of cervicodorsal spine with screening of brain was normal (figure 1). Whole body 18 flurodeoxyglucose positron emission tomography scan failed to reveal any abnormal findings. Electrophysiological studies (nerve conduction study, visual evoked potential and brainstem auditory evoked responses) were normal. Lip biopsy carried out to look for pathological features of Sjogren's syndrome was also negative. In view of an episode of lower gastrointestinal bleeding the patient was subjected to colonoscopy and subsequent biopsy. Colonic biopsy findings were consistent with UC.

DIFFERENTIAL DIAGNOSIS

All the routine differential diagnoses for posterolateral column non-compressive myelopathy were considered in the index case, such as nutritional deficiency (vitamin B₁₂, copper), infective (HIV, human T-cell leukemia virus type 1 (HTLV-1)), auto immune (Sjogren's syndrome, systemic lupus erythematosus,) and non-infective inflammatory

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Figure 1 Normal brain MRI T2-weighted images (A) and gadolinium enhanced T1 images (B) T1 sagittal (C), T2 sagittal (D) and T2 fluidattenuated inversion recovery (E) images of MRI of the cervicodorsal spine showing no significant cord pathology; (E) solid arrow pointing towards probable haemangioma in dorsal vertebrae D8.

(sarcoidosis). However, all the relevant investigations were negative except for positive antinuclear antibody and positive CSF oligoclonal bands indicating an immune-mediated pathology.

TREATMENT

In view of evidence of a concomitant inflammatory disease (UC), temporal correlation between the myelopathy symptoms and inflammatory bowel disease with lack of definitive evidence for alternative aetiology on diagnostic workup, the non-compressive myelopathy in our patient was attributed to an extraintestinal manifestation of UC. As infective causes were ruled out, she was started on a course of intravenous methyl prednisolone (1000 mg/day for 5 days) followed by oral prednisolone (60 mg/day) for 6 weeks. In addition, she was initiated on oral mesalamine (2.4 g daily per orally; delayed release preparation) for inducing remission of her UC with subsequent maintenance therapy.

OUTCOME AND FOLLOW-UP

The patient noted tremendous improvement in her symptoms, with ataxia and weakness improving dramatically at 1 month follow-up. Steroids were tapered over the next 8 weeks and at present she is on alternate day maintenance dose (5 mg orally). At 2 months follow-up, bladder symptoms with loss of dexterity of hands and feet remain the only significant residual deficits. No further episodes of bloody diarrhoea were observed. The initial response of the diarrhoeal episode to antimicrobials suggests an infective aetiology to be responsible for precipitation of UC in our patient.

DISCUSSION

Extraintestinal manifestations of UC are relatively common with a lifetime risk of developing at least one of them being 50%.¹ However, neurological extraintestinal manifestations (central as well as peripheral) are relatively uncommon, with the majority of them being documented as isolated case reports only. The neurological extraintestinal manifestations of UC might be attributed to primarily being due to either the disease process per se (autoimmune mediated, thromboembolism) or secondary pathology (infections secondary to immunosuppression, medication related side effects, malabsorption related vitamin deficiencies and toxic/metabolic insults).¹⁻³ Peripheral neuropathy (sensorimotor/autonomic; demyelinating and/or axonal) is considered to be the most frequent neurological disorder in

UC.⁴ Sensorineural hearing loss (immune mediated vestibuloacoustic neuropathy) has also been reported.⁵ Patients with UC are at a higher risk of developing thromboembolic cerebrovascular events, which is hypothesised to be secondary to either a hypercoagulable state or cerebral vasculitis.⁶ Myelopathy presenting as slowly progressive spastic quadriparesis without sensory level constitutes an extremely uncommon extraintestinal manifestation of UC.^{1 2} The exact aetiopathogenesis of myelopathy in UC seems to be speculative with nutritional (vitamin B₁₂ deficiency), infective (HTLV-1) and immune mediated process being considered to be the potential putative mechanisms.¹ Neuroimaging studies are mostly unremarkable. Non-compressive myelopathy as the presenting manifestation of UC has been reported only once before in the literature,² with the neurological symptoms preceding the gastrointestinal manifestations by 4 months. Our patient was unique in that the presenting feature of UC was in the form of a posterolateral spinal column disease, which has not been reported before in the literature. On initiation of steroids, significant symptomatic improvement was observed in our patient. In view of lack of definitive guidelines for the evaluation and management of neurological extraintestinal manifestations of UC, our case understates the need for extensive evaluation in order to rule out the other more common aetiology of posterolateral spinal column disease (nutritional (vitamin B₁₂, copper deficiency), infective (HTLV-1, HIV), autoimmune diseases (Sjogren's disease)), prior to incriminating immune mediated process related to UC as the potential aetiopathogenic factor.

Learning points

- ▶ Non compressive posterolateral column myelopathy may pose a stiff diagnostic challenge in many cases.
- ▶ Immune-mediated diseases such as ulcerative colitis may rarely manifest as a posterolateral column non-compressive myelopathy.
- ▶ Ulcerative colitis should be considered in the aetiological differentials for posterolateral spinal cord column disease in the appropriate clinical scenario.
- ▶ If promptly diagnosed, significant recovery can be achieved, as exemplified in our case.

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Competing interests None.

Patient consent Obtained.

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REFERENCES

- 1 Scheid R, Teich N. Neurologic manifestations of ulcerative colitis. *Eur J Neurol* 2007;14:483–92.
- 2 Lossos A, River Y, Eliakim A, *et al.* Neurologic aspects of inflammatory bowel disease. *Neurology* 1995;45:416–21.
- 3 Loftus EV Jr. Inflammatory bowel disease extending its reach. *Gastroenterology* 2005;129:1117–20.
- 4 Gondim FA, Brannagan TH IIIrd, Sander HW, *et al.* Peripheral neuropathy in patients with inflammatory bowel disease. *Brain* 2005;128:867–79.
- 5 Nemoto H, Iguchi H, Ichikawa Y, *et al.* Ulcerative colitis presenting as sensorineural hearing loss, brain stem encephalopathy and white matter lesions. *Neurologist* 2004;10:165–8.
- 6 Twig G, Zandmann-Goddard G, Szyper-Kravitz M, *et al.* Systemic thromboembolism in inflammatory bowel disease: mechanisms and clinical applications. *Ann N Y Acad Sci* 2005;1051:166–73.

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