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Case Report

Myopericarditis as an Extraintestinal Manifestation of Ulcerative Colitis: A Case Report and Literature Review

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Abstract

Introduction: Extraintestinal manifestations of inflammatory bowel disease (IBD) occur in many patients. Cardiac inflammation in the form of pericarditis, myocarditis and myopericarditis have been described to occur rarely as an extraintestinal manifestations of IBD.

Case Description: We describe a case of a 28 year old male admitted to hospital with a several week history of bloody diarrhea, admitted with a severe Ulcerative Colitis (UC) flare. This patient subsequently developed positional, pleuritic chest pain associated with an EKG showing mild inferolateral ST-segment elevation, an echocardiogram showing mild inferolateral hypokinesis and an elevated troponin—felt to represent myopericarditis. These symptoms completely resolved with corticosteroid treatment, and after a thorough workup was felt to be most likely secondary to the UC flare.

Discussion: Myopericarditis is a rare but very important extraintestinal manifestation of IBD to be aware of, as early identification and treatment can have significant impacts on patient morbidity and mortality.

Keywords: Ulcerative Colitis; Myopericarditis; Extra-intestinal; Case Report

Abbreviations

UC: Ulcerative Colitis; IBD: Inflammatory Bowel Disease; CRP: C-reactive Protein; ECMO: Extracorporeal Membrane Oxygenation.

Introduction

Ulcerative colitis (UC) is an Inflammatory Bowel Disease (IBD) characterized by recurrent episodes of inflammation in the mucosa of the colon, often extending from the rectum proximally in a continuous manner. Extraintestinal manifestations of IBD occur in many patients, from the gastrointestinal mucosa triggering an immune response in extraintestinal sites, due to common epitopes in genetically susceptible individuals [1].

Myocarditis, pericarditis, and myopericarditis are characterized by inflammation of the myocardium, pericardium and both, respectively. These conditions can be triggered by infections, inflammatory conditions, radiation and drugs. This spectrum of cardiac inflammation has been described to rarely occur as an extraintestinal manifestation of UC and has also been reported to be associated with certain IBD therapies [2-6]. However, in a Danish cohort study of 15,572 IBD patients followed over 15 years, the incidence of myocarditis was only 0.03%. In its mild form, this syndrome can cause symptomatic distress, however at its worst can lead to heart failure and death. Whilst rare in incidence, recognition of this syndrome is essential given its potential for significant morbidity.

Case

The patient of focus is a 28-year-old male with a 14-month history of ulcerative pancolitis. Initially he was treated with steroid-induced remission, and subsequently started and maintained on infliximab monotherapy. He was in clinical remission with endoscopic healing seen on subsequent colonoscopy. He was never treated with mesalamine (5-aminosalicylate). He had no cardiac risk factors and took no other medications.

He presented to hospital with a two-week history of progressive bloody diarrhea and one day of chest pain. He described having traveled to on a 4 hour flight to Houston, Texas for leisure when

he began passing up to 10 bowel movements per day with associated left lower quadrant abdominal pain. One day after his 4 hour return flight, he developed progressively worsening pleuritic, positional retrosternal chest pain. He denied radiation of the pain, hemoptysis or any environmental exposures during his trip. The physical examination revealed normal vital signs, heart and breath sounds with no calf swelling or tenderness. The abdomen was tender to palpation in the left lower quadrant without peritonitis.

His hemoglobin was 94 g/L. His stool was tested for bacterial culture, virology, and Clostridium difficile, however all returned negative. Fecal calprotectin was elevated to 1910 mg/kg (normal <50 mg/kg). Flexible sigmoidoscopy demonstrated inflammation with severe colitis and deep ulceration, erythema and friability as well as pseudopolyposis seen from about the descending colon all the way down to the rectum contiguously. (See Figure 1) Pathology was consistent with chronic active colitis. The clinical picture was consistent with a severe ulcerative colitis (UC) flare.

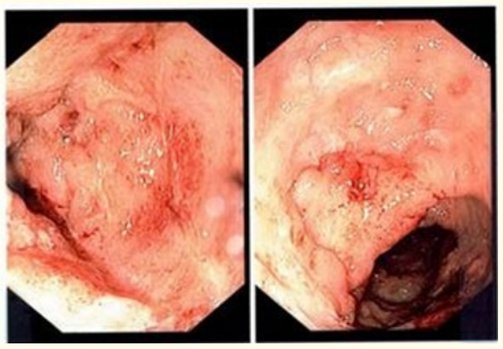

Figure 1: Endoscopy showing acute colitis.

His EKG on admission showed mild inferolateral ST elevation without reciprocal changes, (See Figure 2) with a troponin elevated to a peak value of 66 ng/L (normal less than or equal to 30 ng/L). C-reactive protein (CRP) was elevated at 77.5 mg/L (normal less than or equal to 5 mg/L). D-dimer was 330 µg/L (normal less than 500 µg/L), and given the low clinical suspicion, pulmonary embolism was felt to be unlikely. Subsequent echocardiogram showed an ejection fraction of 54%, no pericardial effusion observed, however demonstrated mildly decreased myocardial deformation of the basal inferior and inferolateral segments.

At this point the clinical picture of chest pain was most consistent with myopericarditis given the clinical symptomatology of pericarditis with an elevated troponin. Epstein Barr Virus serology showed reactive VCA IgG, reactive EBNA but negative EA IgG, suggestive of past infection. CMV IgG antibodies positive with an undetectable viral load, also suggestive of past infection. It was felt that this myopericarditis occurred as a result of the UC flare.

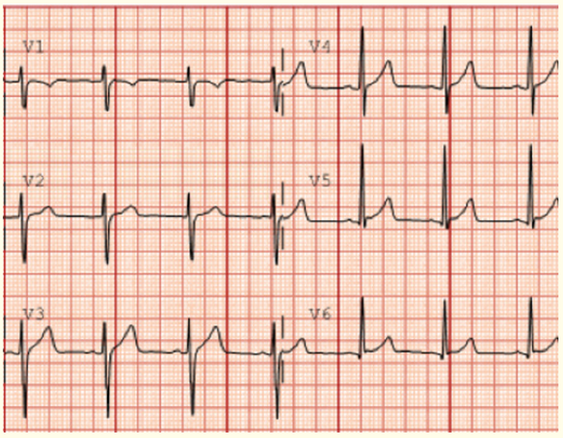

Figure 2: EKG showing ST elevation.

On his second day of hospitalization, the patient also developed right gluteal pain, with HLA-B27, antinuclear antibody and rheumatoid factor screens negative. Subsequent MRI of the sacroiliac joints showing increased T2 signal along the anterior margin of the right SI joint with a small amount of asymmetric soft tissue edema, however no osteitis or ankylosis was observed. This was felt to most likely be an inflammatory bowel disease-related arthritis.

The patient was subsequently started on intravenous methylprednisolone and by the fifth day of treatment had marked improvement chest pain, right gluteal pain and diarrhea, with biochemical improvement in his CRP to 12.4 mg/L. He was discharged home with a prednisone taper, and seen in follow-up two weeks later with clinical stability improved and repeat CRP at 5.5 mg/L. It was felt that this clinical picture was a result of a secondary loss of response to infliximab, and in collaboration with the patient the decision was made to transition to vedolizumab for maintenance IBD therapy.

Discussion

Inflammatory bowel disease can present with a wide variety of cardiovascular complications such as: pericarditis, myocarditis and arrhythmias [12,13]. Pericarditis represents the most common cause of cardiac extra intestinal manifestation of IBD at 70% with a higher prevalence in male ulcerative colitis patients [0.23%) compared to Crohn's disease [0.19%) [14-16].

While myocarditis as an extra-intestinal manifestation of ulcerative colitis is rare [10], the clinical course it takes may vary. Complications ranging from uncomplicated hospitalization to severe heart failure and cariogenic shock requiring extracorporeal membrane oxygenation (ECMO) may arise. Treatment and control of the ulcerative colitis flare often correlates with myocardial disease

regression [4]. It is therefore prudent -in patients with ulcerative colitis- to consider cardiac involvement as an extraintestinal manifestation in the appropriate clinical setting and after excluding more common etiologies, as the implications of specifically targeting ulcerative colitis could prove lifesaving, like in the report using infliximab for the patient on ECMO [11].

Direct cytotoxicity during an acute flare could result from exposure to auto-antigens [12]. Lymphocytic infiltration of the myocardium leading to myocyte apoptosis has been hypothesized as the mechanism driving the process as suggested by the case reports demonstrating biopsy proven myocarditis as a result if ulcerative colitis, the majority exhibited a lymphocytic infiltration of the tissue [6,7]. Several reports in the literature have also described mesalamine and infliximab cardiotoxicity as a cause of myocarditis in ulcerative colitis patients [8,9,17]. However, our patient was not on mesalamine and had been stable on infliximab for many months prior to developing these symptoms, making drug-induced causes unlikely.

In this case, the patient had been suffering from an ulcerative colitis flare of increasing severity for several weeks prior to the onset of chest and gluteal pain. Given the temporality of these manifestations in the absence of alternative triggers, as well as the concurrent resolution of the cardiac involvement when steroids were initiated for the ulcerative colitis flare, it seems that the myopericarditis and sacroiliitis were most likely extraintestinal manifestations of IBD.

Conclusion

In conclusion, pericarditis, myocarditis, and myopericarditis can be rare extraintestinal manifestations of inflammatory bowel disease. Prompt recognition and treatment of this syndrome is essential, as this syndrome can lead to significant morbidity and mortality. Further research should focus on predisposing factors and optimal duration of corticosteroid treatment.

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