CASE REPORT

Small bowel infarction in a patient with coeliac disease

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A 40 year old man was admitted with a four week history of intractable diarrhoea and abdominal pain. A clinical diagnosis of inflammatory bowel disease was supported by biopsies of colonic mucosa. There was no response to Mesalazine and over 12 days the patient became critically ill with diarrhoea, hypovolaemia, and peritonism. A laparotomy was performed and 130 cm of infarcted ileum was resected. Extensive investigations excluded thrombophilia and echocardiography excluded intracardiac thrombus. Postoperatively the patient continued to have diarrhoea and he was diagnosed with coeliac disease on the basis of positive antiendomysial and antitissue transglutaminase autoantibodies and duodenal histology. Although there is no proof that mesenteric infarction occurred as a direct consequence of coeliac disease, clinicians should be aware of this possibility.

A 40 year old man was admitted with a four week history of watery diarrhoea and generalised abdominal pain. He reported no vomiting or rectal bleeding, and no extraintestinal manifestations of inflammatory bowel disease (IBD), such as arthralgia or skin rash. He had no medical history other than acne, for which he was taking minocycline long term. He was a non-smoker, and did not consume excessive alcohol or illegal drugs, such as cocaine. Examination revealed generalised abdominal tenderness with hyperactive bowel sounds. There was no evidence of an inflammatory disorder: C reactive protein, < 5 mg/litre; leucocyte count, 6×10^9 /litre (range, $4\text{--}11 \times 10^9$); and no features of systemic inflammatory response syndrome (pulse, 70; respiratory rate, 15; temperature, 36.9°C). Haematology demonstrated thrombocytosis (481×10^9 /litre; range, $150\text{--}350 \times 10^9$ /litre) and deranged coagulation (prothrombin time, 28 seconds; activated partial thromboplastin time, 41 seconds; international normalised ratio, 2.2). Platelet counts remained raised during admission and repeated intravenous doses of vitamin K were needed to maintain a normal international normalised ratio. Stool was positive for *Clostridium difficile* toxin, but culture was negative for this organism and other pathogens. IBD was diagnosed, based on colonic biopsies demonstrating a mixed inflammatory cell infiltrate in the lamina propria with cryptitis and crypt abscesses.

The patient was treated with a course of Mesalazine—steroids were avoided because infection had not been excluded. Two weeks after admission the patient became critically ill, with shock and generalised peritonism. A computed tomography scan of the abdomen demonstrated an ischaemic small intestine. At surgery, superior mesenteric artery thrombosis was found and 130 cm of gangrenous ileum was resected.

Postoperatively, the patient continued to have diarrhoea. A coeliac disease (CD) screen demonstrated positive antiendomysial and antitissue transglutaminase antibodies. Duodenal

biopsies showed total villous atrophy, pronounced crypt hyperplasia, and large numbers of intraepithelial lymphocytes (IELs) (fig 1A, B). A diagnosis of CD was made. The colonic biopsies were reviewed, and increased numbers of IELs were noted (fig 2A, B). Lymphocytic colitis (LC) associated with CD was diagnosed.

A thrombophilia screen demonstrated a low free protein S concentration and borderline total protein S concentration; this screen was repeated three months later and was normal (table 1). Echocardiography demonstrated a normal heart with no thrombus. A patent foramen ovale could not be excluded, because bubble contrast echocardiography was not available. Electrocardiograms demonstrated sinus rhythm. Antineutrophil cytoplasmic antibodies, anticardiolipin, lupus anticoagulant, antismooth muscle, antimitochondrial, anti-gastric, and antiliver kidney microsomal antibodies were all

Figure 1 (A) Duodenal biopsy (haematoxylin and eosin stained; original magnification, $\times 20$) showing elongated crypts and flattened villi. (B) Duodenal biopsy (haematoxylin and eosin stained; original magnification, $\times 40$) showing increased intraepithelial lymphocytes.

Abbreviations: CD, coeliac disease; IBD, inflammatory bowel disease; IEL, intraepithelial lymphocyte; LC, lymphocytic colitis

Figure 2 (A) Colonic biopsy. (B) Colonic biopsy with increased intraepithelial lymphocytes. Haematoxylin and eosin stained; original magnification, $\times 20$.

negative. Rheumatoid factor had a titre of < 22 and antinuclear antibody was very weakly positive at 1/40. Human immunodeficiency virus and hepatitis serology were negative. The patient's diarrhoea improved with gluten exclusion, and he was discharged to outpatient care.

DISCUSSION

Here, we report mesenteric infarction in a patient with a diagnosis of CD and LC. IBD is causally associated with mesenteric infarction, and a diagnosis of Crohn's disease or ulcerative colitis would explain the association of mesenteric infarction and CD.¹ However, the combination of increased IELs ($> 20/100$ epithelial cells) with cryptitis and crypt abscesses in the colonic biopsies is more typical of LC than IBD,² because 38% of patients with LC have cryptitis and abscesses,² whereas increased IELs are not associated with IBD.³ The absence of ulceration and mucosal abnormalities on endoscopy, with no increases in inflammatory markers such as C reactive protein, also favours LC.² The association of mesenteric infarction and CD might occur by ischaemia "unmasking" CD.³ However, there was no evidence of intestinal ischaemia on admission⁴ (normal abdominal x ray, amylase 46, D-dimers negative, no metabolic acidosis, leucocyte count of 6×10^9) and CD serology was performed on blood drawn before the diagnosis of mesenteric infarction.

"The combination of increased intraepithelial lymphocytes with cryptitis and crypt abscesses in the colonic biopsies is more typical of lymphocytic colitis than inflammatory bowel disease"

The cause of mesenteric infarction in this patient is unclear. The thrombocytosis may have played a role in the mesenteric infarction.^{5 6} It is interesting to note the dramatic

Table 1 Thrombophilia screen

Measure (range)	At presentation	3 month follow up
Free protein S (51–140)	36, low	54
Total protein S (64–166)	65	73
Protein C (74–128)	86	148, high
AT III (83–128)	64, low	111
APC resistance (0.68–1.23)	0.77	0.91
Factor V deficit (0.84–1.25)	0.93	0.97
Factor V Leiden	Absent	Absent
PT 20210A	Absent	Absent

APC, activate protein C; AT III, antithrombin III; PT, prothrombin.

Take home messages

- We report a case of mesenteric infarction in a patient with confirmed coeliac disease
- Although there is no proof that mesenteric infarction occurred as a direct consequence of coeliac disease, clinicians should be aware of this possibility

fall in platelet counts that occurred at the (presumed) time of infarction. The low concentrations of the natural anti-coagulant protein S may have contributed to mesenteric thrombosis.⁷ Protein S is vitamin K dependent, and the vitamin K deficiency in this patient, which presumably resulted from malabsorption, may have produced a reversible protein S deficiency.⁷ Such a mechanism was associated with portal vein thrombosis in a patient with CD reported by Thorburn *et al.*⁸

Two cases of mesenteric infarction in CD have been reported.⁹ However, neither met contemporary diagnostic criteria for CD, thrombophilias were not excluded, and a response to gluten exclusion was not demonstrated.⁹ Here, mesenteric infarction occurred in a patient with CD, with no other cause being identified. Although this case does not prove that mesenteric infarction occurred as a direct consequence of CD, it should alert clinicians to this possibility, and to the potential for CD to cause postoperative diarrhoea.

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