Case Report

A 25-year old Ghanaian woman presented with a 1-week history of fatigue and breathlessness. Menstrual and dietary histories were normal, there was no overt gastrointestinal blood loss, and she took no medication. Physical (including digital rectal) examination was normal, apart from pale conjunctivae. Investigations revealed haemoglobin 4.6g/dL, white cell count 7.09 x 109/L, platelet count 442 x 109/L, MCV 63fL, MCH 16.0pg, serum ferritin 2.0µg/L. Other routine laboratory investigations, including serum anti-tissue transglutaminase, hemoglobinopathy screen, serum haptoglobin and reticulocyte count, were normal. Chest X-ray was normal. After transfusion of 4 units of red cells, the hemoglobin rose to 10.0g/dL with resolution of symptoms, and she was discharged on oral iron.

Soon afterwards, latent tuberculosis was diagnosed after routine screening revealed a positive Quantiferon test, and she was treated for 3 months with rifampicin. Several further transfusions were required during the next five months, the hemoglobin falling at one point to 5.8g/dL. Screening for coagulopathy and Von Willebrand disease was negative, and gastroscopy and colonoscopy were normal, as was duodenal histology. After referral to the gastroenterology service, she underwent capsule endoscopy, which showed fresh blood in the mid-jejunum, with clot and altered blood more distally, but no obvious mucosal abnormality. Computed tomography of the abdomen was unremarkable. Double-balloon enteroscopy revealed an actively bleeding submucosal lesion 1.6m beyond the pylorus, the overlying mucosa appearing intact (Figure 1, upper panel). At laparotomy, a small intestinal polypoidal tumor was resected and histology revealed a lymphangioma (Figure 2, lower panel). She made a good recovery and remains symptom-free with a normal blood count.

Figure 1: Upper Panel: Polypoidal lesion in jejunum during double-balloon enteroscopy

Figure 2: Lower Panel: Resected lymphangioma, showing dilated thin-walled channels covered by small intestinal mucosa (hematoxylin and eosin, x50 magnification).
Lymphangiomas are benign lesions of vascular origin with lymphatic differentiation and usually present in childhood, adult cases being rare. Only a minority (<5%) occur in the abdomen and are often incidental findings during endoscopy or imaging studies [1]. Mesenteric lymphangiomyomas may be large, causing abdominal distension, [2] or small intestinal obstruction secondary to volvulus, [3,4] but rarely cause significant intestinal blood loss. Our case is therefore unusual, and we are aware of only one other case of small intestinal lymphangioma diagnosed by enteroscopy, using the single-balloon technique [5]. Patients with recurrent iron deficiency anemia often present to specialties other than gastroenterology, and after a normal gastroscopy and colonoscopy, capsule endoscopy, followed if necessary by either single- or double-balloon enteroscopy, should be used undertaken to exclude more common pathologies such as Crohn’s disease, tuberculosis, and radiation damage.

References