Case Report

An unusual multiplex cause of severe gastrointestinal bleeding in a haemodialysed patient

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Introduction

Gastrointestinal bleeding is a frequent and sometimes life-threatening complication of end-stage renal disease. The most important causes of bleeding are vascular malformations, peptic ulcers, erosive gastritis and oesophagitis, colonic and rectal ulcers, and diverticular disease [1]. In the spectrum of these abnormalities, vascular malformations have a very specific place, because diagnosis often is difficult and treatment is frequently frustrating [2,3].

Case

A 77-year-old female patient had a history of hypertension since 1966 and according to past medical history, she had not been treated optimally. End-stage renal disease was diagnosed in January 1998. Blood pressure was 200/100 mmHg, serum creatinine 441 μmol/l, urea nitrogen 34.7 mmol/l, creatinine clearance 9.8 ml/min, and serum potassium 6.3 mmol/l. The patient had severe metabolic acidosis. The suspected renal diagnosis was hypertensive nephrosclerosis. Chest X-ray showed cardiac enlargement, ECG showed left ventricular hypertrophy and the patient had evidence of diffuse atherosclerosis. Both kidneys were small (87 × 29 mm and 94 × 32 mm). Haemodialysis was started in January of 1998. Since that time, severe anaemic episodes occurred on seven occasions (lowest haematocrit 10.7%, haemoglobin 3.6 g/dl, and serum iron 2.7 μmol/l), which required multiple blood transfusions and was resistant to iron supplementation, human recombinant erythropoietin, and anti-ulcer drug treatment. She required a total of 29 units of packed red blood cells over a 4-month period. The investigations excluded any coagulation abnormalities, haematological and gynecological diseases. Although there was no evidence of manifest bleeding, the patient’s stool was consistently positive for occult blood.

On thorough investigation of the patient’s gastrointestinal tract, oesophagogastroduodenoscopy showed some erosions in the corpus of the stomach and slightly protruding, parallel longitudinal antral streaks converging on the pylorus, which contained visible tortuous ectatic vessels (watermelon stomach) (Figure 1) and two angiodysplastic lesions in the postbulbar duodenum. Multiple biopsies were performed. Histological examination of the antral biopsy specimen showed a moderately atrophic mucosa with ectatic superficial capillaries. Colonoscopy revealed diffuse atrophic changes of the intestinal mucosa with several

Fig. 1. Endoscopic picture of watermelon stomach showing parallel longitudinal antral stripes of tortuous ectatic vessels converging on the pylorus.
angiodysplasias, 5–7 mm in diameter (Figure 2) and some diverticuli in the sigmoid colon. Because of the multiplicity of the vascular malformations we decided to treat the patient with an oestrogen–progesterone combination (0.05 mg oestradiol and 1 mg norethisterone acetate daily), given orally. Since the initiation of this therapy the patient has not required any further transfusions and her haematological status has remained stable during the subsequent 12-month observation period. No significant side-effects have been noted.

Comment

Significant bleeding from vascular malformations is an important factor of morbidity in patients with end-stage renal disease. These lesions play a role in up to 24% of initial episodes of upper gastrointestinal bleeding and in 53% of recurrent haemorrhage [3]. Angiodysplasias are hereditary or degenerative lesions of dilated, thin-walled vessels. They are relatively common in the elderly and are particularly common in some diseases, including renal failure. In some cases angiodysplasias are asymptomatic, but they can cause occult, chronic or sometimes acute life-threatening bleedings. The lesions are frequently diffuse. Our patient had several angiodysplasias in the duodenum and in the sigmoid colon. This raised the suspicion of additional angiodysplasias in the uninvestigated part of the gastrointestinal tract.

On the evaluation of the patient, another type of vascular malformation, the watermelon stomach, was found. This lesion is characterized by prominent stripes of tortuous, ectatic vessels in the antral region of the stomach, similar to the stripes on a watermelon [5]. Watermelon stomach is a specific type of vascular malformation and this is now a distinct clinical entity within the spectrum of upper gastrointestinal mucosal vascular abnormalities [5]. Chronic gastrointestinal bleeding occurs in more than half of the patients with watermelon stomach. The most important disease groups concomitant with this lesion are autoimmune diseases (mostly systemic sclerosis), chronic renal failure, liver cirrhosis, and chronic congestive heart failure. In the largest series of 45 patients with watermelon stomach six had renal failure [6].

The therapeutic approach in case of bleeding from a vascular malformation depends on the accessibility of the bleeding source. Treatment modalities of angiodysplasias and watermelon stomach are similar. The treatment of first choice generally is an endoscopic method: bipolar or multipolar electrocoagulation, heat probe coagulation and laser or argon plasma coagulation. In cases in whom endoscopic methods are unsuccessful although lesions arecircumscribed, surgical intervention is needed [7]. Unfortunately, operative morbidity and mortality are high in uraemic patients. However, endoscopic and surgical treatment modalities are ineffective when the vascular lesions are spread diffusely over the gastrointestinal tract or when lesions are not accessible for treatment. There are some reports on successful treatment with different types of drugs (hormone therapy, tranexamic acid), among which the combination of oestrogen–progesterone appears to be most effective [4,8,9]. Case reports support the efficacy of oestrogen–progesterone treatment, which is particularly effective in patients with renal failure [9]. In the only double-blind placebo-controlled cross-over trial of patients with normal renal function, oestrogen–progesterone significantly decreased transfusion need from 10.9 to 1.1 units of packed red blood cells [10]. The exact mechanism of the action of this type of therapy is still not known. In dialysis patients the bleeding time shortens after the administration of oestrogens [11,12]. It has been shown that these compounds improve the integrity of the vascular endothelium and reduce prostacyclin production [4]. Some reports showed that the duration of their beneficial action on bleeding lasts longer than the administration of the drug [12]; therefore a reduced dose therapy (administration every second or third day) or an intermittent therapy may also be effective.

Side-effects are minor or non-existent. In postmenopausal women the potential side-effects are a slightly increased risk of endometrial and breast cancer, an increased risk of thromboembolism, headache, bloating, and unpredictable uterine bleeding. On the other hand, hormone replacement therapy reduces the risk of ischaemic heart disease, osteoporosis, and fractures, and thus increases life expectancy. In premenopausal women, the effects are similar to those of oral contraception. For males the long-term effects of hormone therapy are less well defined, the most important side-effects being gynecomastia, loss of libido, erectile dysfunction, and penile or testicular atrophy [4].

The case presented here is unique, in that the source of the severe, prolonged gastrointestinal blood loss...
was caused by two types of vascular malformation, namely watermelon stomach and multiple angiodysplasias along the gastrointestinal tract. Our case also demonstrates the excellent efficacy of oestrogen–progesterone treatment in an elderly woman with chronic renal failure on haemodialysis.

Two major lessons can be learnt from this case report

(i) Bleeding from vascular malformations is a significant factor of long-standing, erythropoietin-resistant anaemia of patients with end-stage renal disease. Exact diagnosis is often difficult and requires thorough investigation of the patient.

(ii) Oestrogen–progesterone treatment should be the first treatment of choice for multiple bleeding vascular malformations of the gastrointestinal tract in uraemic patients in case of high transfusion need, when these malformations are inaccessible or refractory to endoscopic therapy and the risk of operation is unacceptably high or surgical intervention is refused.

References


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