CASE REPORT

Pseudomelanosis duodeni in a postrenal transplant patient

Theresa Schuerle, Elie Aoun, Kofi Clarke

West Penn Allegheny Health System, Pittsburgh, Pennsylvania, USA

Correspondence to

Dr Elie Aoun,
eaoun@wpahs.org

SUMMARY

Pseudomelanosis duodeni is a rare entity characterised by dark pigmented intracellular granules seen within macrophages that lie within the lamina propria of the duodenal villi. There is no known treatment, and the clinical significance and long-term sequelae of this entity are unclear. We present a case of *pseudomelanosis duodeni* in a 54-year-old woman who presented with a 1-month history of nausea, vomiting and non-bloody diarrhoea. The medical history was significant for diabetes mellitus type 2, end-stage renal disease status postkidney transplant, hypertension, anaemia of chronic disease and hypothyroidism. A gastroduodenal endoscopy revealed pigmented dark lesions in the duodenal mucosa. Biopsies from the second part of the duodenum and duodenal bulb showed pigmented macrophages in the lamina propria. The findings were consistent with duodenal melanosis. In spite of renal transplant with normalisation of renal function, the duodenal melanosis persists, which raises questions on the role of renal impairment in this entity.

BACKGROUND

This case report highlights a rare entity, *pseudomelanosis duodeni*, which may pose a diagnostic challenge to clinicians who are unaware of the condition, and subsequently may lead to extensive and expensive workup. In addition, the persistent pigmentation postrenal transplant raises questions on the true role of chronic kidney disease in this entity.

CASE PRESENTATION

A 54-year-old woman presented with a 1-month history of nausea, vomiting and diarrhoea. The medical history was significant for diabetes mellitus

type 2, end-stage renal disease status postkidney transplant in 2006, hypertension, anaemia of chronic disease and hypothyroidism. Diarrhoea was described as five watery, non-bloody stools per day. She denied abdominal pain. Her medications consisted of insulin, pantoprazole, hydralazine, losartan, diltiazem, paroxetine, thyroxine, atorvastatin, cyclosporine, mycophenolate mofetil and trimethoprim/sulfamethoxazole. The patient denied tobacco, alcohol or illicit drug use.

INVESTIGATIONS

The stool culture, ova and parasite, leucocytes and *Clostridium difficile* toxins A and B were all negative. A gastroduodenal endoscopy revealed pigmented dark lesions in the duodenal mucosa (figure 1). Biopsies taken from the second part of the duodenum and duodenal bulb showed pigmented macrophages in the lamina propria on H&E stain (figure 2). Staining of the specimen with Masson-Fontana stain was positive, consistent with melanosis (figure 3). Periodic acid-Schiff stain (PAS) and iron staining with Perls' stain were negative (figure 4).

DIFFERENTIAL DIAGNOSIS

The findings were consistent with melanosis in the duodenum.

OUTCOME AND FOLLOW-UP

Colonoscopy performed 2 days later for diarrhoea was normal, and random biopsies of the colon showed benign colonic mucosa without signs of active inflammation or evidence of melanosis coli (figure 5).

The patient's symptoms improved after her immunosuppressive regimen was adjusted and the dose of mycophenolate mofetil was reduced.

Figure 1 Duodenal mucosa with pigmented dark lesions seen on upper endoscopy.

Figure 2 H&E staining of duodenal mucosa— $\times 40$ magnification.

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Figure 3 Masson-Fontana staining of duodenal mucosa— $\times 40$ magnification.

A well-known side effect of mycophenolate mofetil is diarrhoea. The temporal relationship between an improvement in diarrhoea and the reduction in dosage makes it clinically likely that mycophenolate mofetil was responsible for diarrhoea. She continues to remain clinically well, is tolerating her immunosuppressive regimen and has had no signs of allograft rejection.

DISCUSSION

Pseudomelanosis duodeni is a rare entity characterised by dark pigmented granules seen inside the macrophages that lie within the lamina propria of the duodenal villi. It appears to be more common in women in the sixth to seventh decades of life and has distinctive endoscopic, histopathological and electron microscopic features.¹ First described in 1976, it was initially thought to be a form of stored iron.² *Pseudomelanosis duodeni* has also been described in patients with hypertension, chronic renal failure, chronic heart failure and diabetes mellitus.^{1 3–5}

There are several proposed mechanisms on pathogenesis, and no clear aetiology has been described. It has been suggested that melanosis could result from iron deposition in the duodenal mucosa secondary to intramucosal haemorrhage or impairment of luminal iron transport after oral iron supplementation.^{1 6} This theory is supported by electron microscopy that shows ferrous sulfide in the pigment.⁷ It has also been speculated that the pigment may be a combination of melanin-like substances, haemosiderin, lipomelanin and lipofuscin.^{1 6} Other reports have noted an association between *pseudomelanosis duodeni* and certain drugs, including propranolol, hydralazine, hydrochlorothiazide and furosemide.^{1 3 4}

Figure 4 Iron staining of duodenal mucosa— $\times 40$ magnification.

Figure 5 Normal colonic mucosa seen on colonoscopy.

The patient had diabetes mellitus, a history of end-stage renal disease and hypertension which have been described as associations in previous case reports. The patient had persistent duodenal melanosis postrenal transplant with normalisation of kidney function. This supports the assertion that the condition is associated with kidney disease, but perhaps not caused by kidney disease. The patient improved after the immunosuppression regimen was adjusted. Her symptoms were not directly related to duodenal melanosis, but it may have been associated with the medication hydralazine as has been reported previously.

There is no known treatment, and no specific follow-up protocols have been outlined in previously described cases. The clinical significance and long-term sequelae of this entity are also unclear. The darkened endoscopic appearance is unusual and may pose an initial diagnostic challenge to clinicians who are unaware of this condition.

Learning points

- ▶ *Pseudomelanosis duodeni* is a rare entity characterised by dark pigmented granules seen inside the macrophages that lie within the lamina propria of the duodenal villi.
- ▶ Although no clear aetiology has been described, there are several proposed mechanisms on pathogenesis, including iron deposition in the duodenal mucosa secondary to intramucosal haemorrhage or impairment of luminal iron transport after oral iron supplementation; certain drugs, including propranolol, hydralazine, hydrochlorothiazide and furosemide, have also been associated with *pseudomelanosis duodeni*.
- ▶ There is no known treatment and no specific follow-up protocols for *pseudomelanosis duodeni*.
- ▶ The clinical significance of *pseudomelanosis duodeni* is unclear and poses a diagnostic challenge for clinicians.

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