Pseudomelanosis intestini “from pylorus to jejunum:” A rare endoscopic finding in a patient with GI bleeding

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Abstract

Pseudomelanosis of the gastrointestinal (GI) tract is a rare condition used to describe the accumulation of pigment deposits in the intestinal mucosa. Its underlying cause is not well understood. It has been described in association with gastrointestinal hemorrhage, chronic kidney disease, hypertension, diabetes mellitus, and medications such as hydralazine, ferrous sulfate, and furosemide. Melanosis coli is a well-known condition associated with the use of anthranoid laxatives; however, pseudomelanosis of the small intestine is extremely rare and most commonly described in the duodenum, with few cases in the gastric mucosa and even more rare in the jejunum. Herein, we report a case of pseudomelanosis intestini involving the pylorus, duodenum, and proximal jejunum in a patient presented with GI bleeding. The clinical significance of this condition is unknown; however, gastroenterologists should be aware of its existence.

Keywords: Gastro-duodeno-jejunum, gastrointestinal pigmentation, pseudomelanosis

Introduction

Gastrointestinal melanosis is a rare condition described first by Bosordi and Kleinman in 1976 in reference to the endoscopic appearance of black speckled pigmentation of the mucosa.[1] It has been described in association with gastrointestinal hemorrhage, chronic kidney disease, hypertension, diabetes mellitus, and medications such as hydralazine, ferrous sulfate, and furosemide.[2-5] Melanosis coli is a well-known condition associated with the use of anthranoid laxatives; however, pseudomelanosis of the small intestine is extremely rare.[6,7] The underlying cause and the clinical significance of this condition is unknown; however, gastroenterologists should be aware of its existence. Here, we report a case of pseudomelanosis intestini involving the pylorus, duodenum, and proximal jejunum in a patient presented with recurrent GI bleeding.

Case History

A 70-year-old African–American female with past medical history significant for hypertension, chronic obstructive pulmonary disease, chronic kidney disease stage-IIIb, and permanent atrial fibrillation on anticoagulation with warfarin. She presented to the emergency department complaining of recurrent melena. Her home medications included aspirin, clopidogrel, warfarin, hydralazine, bumetanide, metformin, metoprolol, and albuterol inhaler.

On physical examination she was alert, oriented, and not in acute distress. Her vital signs were: Temp 97.8°F; pulse 70 bpm; blood pressure 113/69; and respiratory rate 14 bpm. The abdomen was soft and lax without tenderness, rebound, or organomegaly, auscultation revealed normal bowel sounds, and point of care fecal occult blood test was positive.

Her initial blood workup revealed normocytic hypochromic anemia with a hemoglobin of 6.9 gm/dl (9.2 gm/dL two

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weeks prior to this admission), platelets 229 × 10^9/mcl, and prothrombin time/INR 19.8 seconds/1.8. Serum iron studies revealed a ferritin level of 1704 ng/mL, iron of 74 mcg/dL, total iron binding capacity (TIBC) 183 mcg/dL, unsaturated iron binding capacity (UIBC) 109 mcg/dL, and iron saturation of 15%.

She underwent esophagogastroduodenoscopy (EGD) which revealed one non-bleeding superficial gastric ulcer with a clean base (Forrest class III) on the lesser curvature of the gastric antrum and diffuse duodenal melanosis. She was discharged home after stabilization and packed red blood cells transfusion. She presented again two weeks later with the same complaint of melena, a repeat EGD showed a healed gastric ulcer with no stigmata of bleeding, and diffuse pseudomelanosis of the gastric antrum, pylorus [Figure 1], duodenum and proximal jejunum [Figure 2]. Histopathology of the gastric antrum and duodenum revealed macrophages containing dark pigmented granules within the lamina propria [Figure 3a and b], nonreactive to Perl’s iron stain [Figure 3c], a picture consistent with pseudomelanosis mucosal changes.

She was readmitted three weeks later with similar complaints. An upper and lower device-assisted enteroscopy with fluoroscopy was performed which revealed pseudomelanosis extending throughout the gastric antrum, duodenum, and jejunum (280 cm distal to the pylorus). A 5 mm polyp in the cecum was removed and revealed tubular adenoma, the remaining parts of the colon and terminal ileum were normal with no evidence of melanosis.

**Discussion**

Pseudomelanosis intestini is a benign condition that refers to the rare endoscopic appearance of dark pigment deposits in the intestinal mucosa. The condition has a female predominance and is mostly seen in the 6th and 7th decades of life. It is characterized by the accumulation of pigment-laden macrophages in the lamina propria, and is thought to be secondary to iron sulfide, hemosiderin, or lipomelanin deposition within the lysosomes as suggested by histochemical analysis and electron microscopy.

Pseudomelanosis is generally recognized on endoscopy; however, the appearance of pigmented gastrointestinal mucosa makes this a challenging dilemma as the differential diagnosis includes clinically significant conditions such as metastatic malignant melanoma, brown bowel syndrome, hemosiderosis, and hemochromatosis. True melanosis associated with malignant melanoma does not stain with Prussian blue, but rather would be highlighted with the Masson-Fontana stain, revealing the melanin pigments. However, this stain is not specific for melanin, and other immunohistochemical stains would benefit in the differentiation of malignant melanoma from pseudomelanosis. Brown bowel syndrome involves the deposition of lipofuscin in the intestinal tunica muscularis and usually spares the actual mucosal lining. It is usually seen with certain vitamin deficiencies but it may also be secondary to an underlying intestinal malignancy.

The differentiation between brown bowel syndrome and pseudomelanosis is crucial for identifying the etiology and management of each condition. In hemosiderosis, diffuse strong iron staining of all pigments would be present with the Prussian blue iron stain, in contrast to pseudomelanosis, where there is partial and relatively weak iron positivity. Although pseudomelanosis contains iron components, some biopsies will not stain positive with the Prussian blue iron stain. This discrepancy is thought to be due to iron in the pigments occurring in the iron sulfide form rather than in the iron oxide form, which is not highlighted with the Prussian blue or Perl’s iron stain. This is evident in our patient, in which there was a recent history of gastrointestinal hemorrhage yet biopsy specimens resulted in negative Perl’s iron stain. This finding suggests that the pigmented mucosa is most likely not a result of hemosiderin deposition from the gastrointestinal hemorrhage.

It has been proposed that pseudomelanosis is associated with impaired luminal absorption of iron, as it is most commonly seen in the proximal duodenum, the site of maximal iron absorption. This has been hypothesized to be secondary to coupling of iron with sulfur or cyclic compounds, leading to the accumulation of the pigment within the mucosa, the source of sulfate is unclear; however, antihypertensive drugs such as hydralazine and foods that contain sulfate are thought to be the culprit. Nonetheless, this does not explain the extra-duodenal sites of pigments or the association where sulfur containing ingestions are not found. This case is also unique because the pseudomelanosis pigmentation is not confined to areas in the GI tract with known iron absorption, and the distribution of pigmentation is much greater than most cases reported in the literature.
Another contributing factor that has been hypothesized is a defect in macrophage metabolism that leads to the accumulation of the iron-containing pigments.[17] It has been suggested that this defect is acquired rather than congenital, which is consistent with the chronological appearance of new pigmented mucosa as seen in this patient.

This case illustrates the importance of identifying the etiology of pigmented mucosa in the gastrointestinal tract and the importance of excluding other causes of the pigmentation. Furthermore, the correlation between the site of pigmentation, the extent of involvement, the clinical significance and long-term sequelae have yet to be fully established.

**Conclusion**

The etiologic mechanism, clinical significance, and prognostic and long-term impact of pseudomelanosis intestini remains unknown. Also, there is no established recommendation regarding the need for therapeutic interventions or regular upper endoscopy surveillance.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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**Conflicts of interest**

There are no conflicts of interest.

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