

# CASE REPORT

## Persistent Obscure Gastrointestinal Bleeding After Jejunal Resection of a Bleeding Jejunal Angiodysplasia in a Lady with Multiple Medical Problems

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### ABSTRACT

We report a 49 years old chinese lady with mitral valve replacement on warfarin, beta – thalassemia trait, chronic hepatitis B, diabetes mellitus and gouty arthritis who had iron deficiency anemia and recurrent malaena since 1999. Oesophagogastroduodenoscopy (OGDS), colonoscopy, angiographic studies and the red cell scan did not reveal any source of gastrointestinal bleeding. Capsule endoscopy (CE) revealed a bleeding jejunal mass. It was resected on laparotomy and histopathology reported the mass as angiodysplasia. However she had persistent malaena and symptomatic anaemia with recurrent blood transfusions. Push enteroscopy was normal. Possible reasons for her persistent bleeding included multiple synchronous angiodysplasia, warfarin use, NSAID gastropathy and diastolic heart failure.

**Key Words:** Angiodysplasia, capsule endoscopy, iron deficiency anemia

### Case report

This was a 49 years old chinese lady with metallic mitral valve replacement and atrial fibrillation on warfarin since 1982, thalassemia trait, chronic hepatitis B, diabetes mellitus and gouty arthritis. She presented to us with malaena and iron deficiency anemia since 1999. Her medications included adjusted dose warfarin with an aim of INR between 1.5 to 2.0, frusemide, spironolactone, digoxin, captopril, allopurinol and folate.

She was investigated initially with oesophagogastroduodenoscopy (OGDS) which was normal. Colonoscopy revealed fresh malaenic stool covering the mucosa of her ascending colon. Selective superior mesenteric artery (SMA), inferior mesenteric artery (IMA) and celiac trunk angiography (Figure 1), red cell scan using stannum/technetium 99m in 2001 and

small bowel barium follow-through did not reveal any possible source of bleeding. CT scan abdomen with contrast showed congestive hepatomegaly and engorged inferior vena cava (Figure 2). Transthoracic echocardiography showed that she had dilated heart chambers with moderate tricuspid regurgitation and pulmonary hypertension.

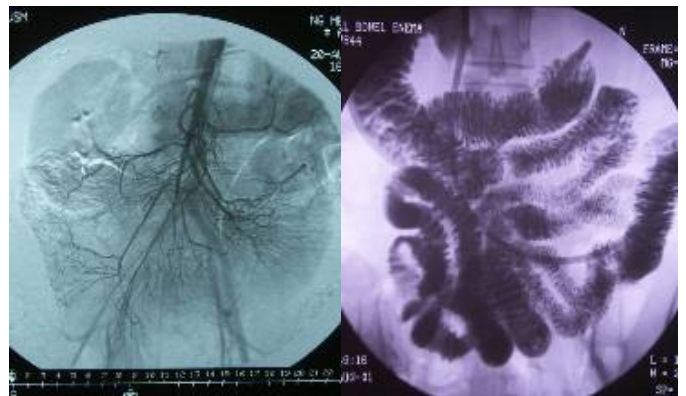


Figure 1. Normal mesenteric angiographic and small bowel enema study.

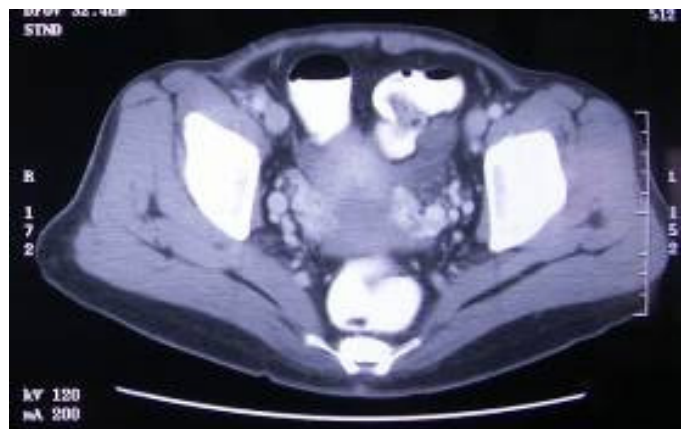


Figure 2. CT scan abdomen showing engorged pelvic veins due to the engorged inferior vena cava.

She was referred to another centre for capsule endoscopy in 2005. There was fresh bleeding seen at the proximal jejunum (Figure 3). Subsequent open laparotomy found a small intestinal mass measuring 2x2cm at proximal jejunum approximately 25cm from the duodeno-jejunal junction which was resected. Histopathology reported the mass as angiodysplasia.

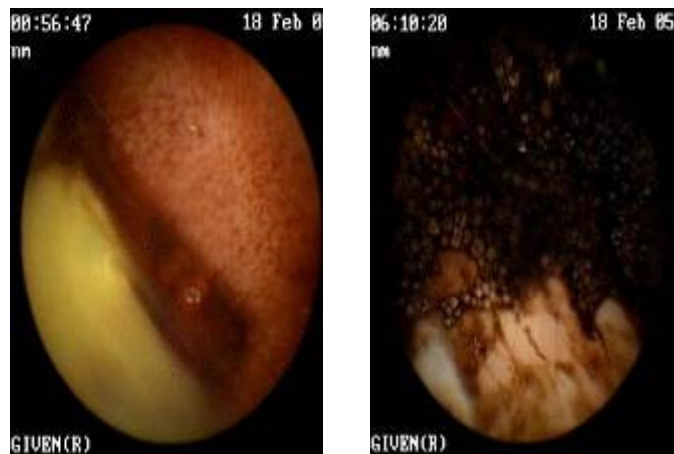


Figure 3. Capsule endoscopy showing bleeding in the jejunum.

She still had recurrent melaena and symptomatic anemia weeks later. A repeat OGDS revealed multiple antral ulcers. This was attributed to the non-steroidal anti-inflammatory (NSAID) agents she took for her gouty arthritis. She was advised to stop the NSAID and a course of proton pump inhibitor was given. A later OGDS showed the antral ulcers had healed. However, she still complained of recurrent melaena and anaemia. She had a push enteroscopy examination in another centre which was normal. She also had a trial of estrogen therapy but failed to keep her from persistent bleeding.

## Discussion

This case posed a both diagnostic and therapeutic dilemma. Angiodysplasia of the gastrointestinal tract was a common cause of obscure or occult gastrointestinal bleeding but angiodysplasia involving the small intestine was a rare clinical entity accounting for only 16% of all angiodysplasia[1].

Small intestinal angiodysplasia can occur at any age. The angiodysplasia has a congenital origin for those <20 years old and it is degenerative in the older age group. Jejunal angiodysplasia is twice more common than the ileal type.

Imaging the small intestine can be very challenging because this area was not reachable for most conventional endoscopic methods including the oesophagogastroscope and colonoscopy. Angiographic studies and red cell scan can miss 80 to 90% of the bleeding (false negative rate of 10 to 20%) because a certain volume of bleeding was required to be detected on angiography or red cell scan[2,3].

Capsule endoscopy (CE) is an effective, safe and non-invasive method to visualize the small intestine. The overall yield of obscure gastrointestinal bleeding identification using capsule was 50-70%[4]. However the capsule was not reliable in excessive bleeding as it obscured the camera and in gastro-

intestinal hypermotility. It also did not allow any biopsy or therapeutic procedure unlike endoscopic methods. Furthermore it was not widely available in Malaysia and few patients can afford it in the private.

Enteroscopy can visualize the small intestine directly with the additional benefit of therapeutic procedures. Push enteroscopy (PE), sonde enteroscopy and intraoperative endoscopy had diagnostic yields of 45 - 80%, 77% and 70% respectively[5]. Few trials had shown that CE however was more superior to PE in detecting causes of obscure gastrointestinal bleeding[5] and probably cheaper.

More recently, double balloon enteroscopy (DBE) was introduced as an alternative to CE with a comparable yield for visualizing the small intestine and the additional advantage for biopsy, to and fro examination of an area of interest and therapeutic procedure[6]. This technique however was not available in Malaysia until recently, expensive and required training with a learning curve.

Capsule endoscopy had identified jejunal angiodysplasia as the cause of bleeding in this patient. Peptic ulcer disease as a result of the non-steroidal anti-inflammatory agent (NSAID) which she took for her gout was not all the answer to her persistent melaena. The warfarin use was another cause of her bleeding. Since she needed the warfarin for her metallic valve, the INR was monitored closely for a balance between the risks of bleeding and valve thrombosis which can be very difficult.

Clouse et al had shown that 60% of their 600 cases had more than one angiodysplasia[7]. Multiple angiodysplasias tend to cluster in similar segments but synchronous lesion was reported in 20%. This lady may have undetected multiple synchronous angiodysplasia elsewhere. A more sensitive examination especially double balloon enteroscopy should be considered but it is limited by its availability and cost. More recently, a case was reported on diffuse microscopic angiodysplasia[8] which cannot be seen endoscopically.

Hemodynamic changes to the small intestinal vasculature as a result of her diastolic heart failure may have predisposed her to the formation of angiodysplasia[9]. Other local factors involved in the pathogenesis of angiodysplasia include intermittent venous obstruction, intermittent arterial flow and local vascular degeneration.

Non-invasive endoscopic treatment using enteroscopy was preferred over surgical methods since she had multiple medical problems which can increase her postoperative risks. Since double balloon enteroscopy was not widely available in Malaysia, only medical therapy can be offered and a trial of hormonal therapy using oestrogen-progesterone preparation was given to this patient. Hormonal therapy was usually the last resort and the few studies available had contradicting results[10,11]. However the response in this patient was poor.

## Conclusion

This patient posed a both diagnostic and therapeutic challenge in managing her recurrent obscure gastrointestinal bleeding. Investigations like capsule endoscopy and double balloon enteroscopy was not easily available in Malaysia and was expensive. The therapeutic challenge was a result of her underlying medical problems which were difficult to manage. The last resort of hormonal therapy had also failed.

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