Arteriovenous Malformation of the Stomach: A Rare Cause of Upper Gastrointestinal Bleeding

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SUMMARY
Haemorrhage arising from gastric arteriovenous malformation (AVM) is rare and normally occurs in the elderly. Bleeding gastric AVM presenting in the younger age group is even rarer. We report a case of a 14-year-old boy who presented with recurrent episodes of haematemesis. He subsequently underwent a proximal gastrectomy and the histological examination confirmed a gastric AVM. After reviewing the literature we believe this is the youngest ever reported case of bleeding gastric AVM reported in English literature.

KEY WORDS:
Arteriovenous malformation, upper gastrointestinal bleeding, stomach

INTRODUCTION
Haemorrhage from upper gastrointestinal tract (GIT) is commonly due to bleeding peptic ulcer disease, oesophageal varices and Mallory-Weiss tear. On the other hand, bleeding from gastric arteriovenous malformation (AVM) is an extremely uncommon occurrence accounting for only 1-2% of upper GIT haemorrhage cases1. In addition, a slow, intermittent bleed makes the diagnosis difficult and challenging. Most reports postulate an acquired degenerative aetiology making the commonest age of presentation to be in the 60s to 80s2. Thus far, the youngest patient reported with gastric AVM was 38 years of age. We present a case of chronic, intermittent upper GIT bleed in a young teenager who was eventually diagnosed to have AVM at the fundus of the stomach.

CASE REPORT
A 14-year-old boy was referred from another hospital for further management after he presented with 3 months history of intermittent fresh haematemesis associated with left hypochondrium pain. The bleeding was minimal (1 to 2ml) and intermittent every 2 to 3 weeks. He was admitted a few times to the previous hospital during which his blood pressure and pulse remained stable while his haemoglobin never dropped below 12g/dL. He was otherwise healthy with no family history of bleeding diathesis.

Extensive investigations had been performed during previous hospital admissions which included oesophagogastroduodenoscopy (OGDS), bronchoscopy, fibroptic nasopharyngolaryngoscopy, Computed Tomography (CT) of abdomen and barium meal and follow-through. All results were normal. He was then transferred to our hospital. During the first hospital admission here, an OGDS done during the acute bleeding episode revealed submucosal haemorrhages at the gastric fundus with blood clots and no active bleeding noted. He was observed for 3 days and later discharged after no further bleeding occurred. Unfortunately he was readmitted 2 weeks later as he developed another episode of haematemesis and repeat OGDS revealed focal areas of inflammation with focal umbilicated ulcer or fissure at fundus of the stomach with contact bleeding. The lesion was coagulated with argon (APC) but he continued to have haematemesis a few days later during the same admission. With the inconclusive endoscopic studies, we decided to proceed with coeliac angiographic study which did not show any abnormality. At this point, differential diagnoses of gastrointestinal stromal tumour (GIST), leiomyoma and vascular malformation were entertained. An endoscopic ultrasonography (EUS) showed a 6-mm submucosal lesion at the fundus of the stomach (Figure 1). With a preoperative diagnosis of GIST, a laparoscopic wedge resection of the lesion was planned.

During laparoscopy, there was difficulty in identifying the lesion even after adequate mobilization of the gastric fundus. This was converted to an upper midline laparotomy.

Direct inspection and palpation of the stomach was normal. An anterior gastrostomy was performed but no abnormalities were seen from within. A proximal gastrectomy with pyloroplasty was decided upon based on the pre operative EUS findings.

Grossly, the specimen showed a few areas of dark brown mucosa on the marked anterior wall with no obvious mass or ulcer noted. Microscopically, there were extensive mucosal and submucosal haemorrhages with increased number of dilated and congested blood vessel consistent with AVM of the stomach (Figure 2). The patient recovered well from the surgery.

DISCUSSION
Angiodysplasia and vascular ectasia are the subsets of arteriovenous malformation (AVM) which constitute the presence of dilated and tortuous vessels3. AVM of the gastrointestinal tract have been found to be the single most
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common cause of chronic and massive haemorrhage when conventional diagnostic tools failed to reveal the cause. It accounts for only 1 - 2% of upper GI haemorrhage. Common site for bleeding AVM is the caecum and ascending colon (77.5%), while bleeding from other sites being quite rare; jejunum (10.5%), ileum (5.5%) and stomach (1.4%) in the present case, we had difficulty making the diagnosis due to the small amount of bleeding and the rare site of bleeding AVM.

The cause of GI AVM is unclear but many had attributed it to the degenerative changes on the wall of the bowel. This makes the disease to be more prevalent among the 60s to 80s age group. AVM occurring at sites other than the colon was believed to be congenital in origin, but the youngest patient reported so far in the English literature is 38 years old. Thus, our patient is the youngest patient currently diagnosed with bleeding gastric AVM.

Upper GI endoscopy remains the first diagnostic investigation for patients with upper GIT bleeding. Locating a gastric AVM can be successfully made by OGDs, but its role is limited in an actively bleeding lesion. Macroscopically it may appear as flat or nodular bright red lesion and sometimes appear in telangiectatic form. Endoscopic ultrasound (EUS) has been proven to accurately detect submucosal lesion. With EUS, AVMs appear as hypoechoic structure in submucosal layer and can be easily differentiated from nonvascular structure using an echoendoscope using colour Doppler. However it might fail to disclose the diagnosis in 22% of patients. As in our case we did not proceed with colour Doppler study thus the vascular nature of the lesion was not confirmed.

Accurate bleeding site is the most important factor in determining the appropriate management of patients with AVMs of the GIT. Moore et al had studied 17 patients with bleeding AVM and found out that most bleeding site can be accurately determined preoperatively by selective angiographic studies. It has a diagnostic accuracy of 50 to 75%, but this approaches 90% if the procedure is performed while the patient is actively bleeding. However the coeliac angiography in our patient failed to reveal any abnormalities and this was probably due to the intermittent and small amount of bleeding from the AVM.

Whilst CT and Magnetic Resonance Imaging (arteriography) MRI (A) had their established role in diagnosis and management of cerebral AVM, their role in evaluating GI AVM is still not of much value. CT in our case was performed to look for bowel related masses as the cause of bleeding. Close long term follow up is important in patient with AVM as post operative bleeding ranges from 5% to 37%. This is secondary to incomplete initial resection and/or metachronous AVMs which have been reported in 11% of cases. Our patient had been followed up for 3 months with no recurrence of bleeding.

In summary, gastric AVM should be considered as a rare cause of upper GI bleed in cases where no other abnormalities can be detected. Diagnosis however can be extremely difficult as they may present with a small intermittent bleed as illustrated in our patient. Our patient was the youngest ever reported with such a problem in the English literature.

REFERENCES

Figure 1: Endoscopic ultrasonography showing submucosal lesion (arrow).

Figure 2: Section shows oedematous submucosa with abnormal, dilated and congested blood vessels within.