A belly of blood: A case report describing surgical intervention in a gastric intramural haematoma precipitated by therapeutic endoscopy in an anticoagulated patient

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ABSTRACT
Gastric intramural hematoma, “intramural dissection” or “false aneurysm”, is a rare and dangerous condition which may be more broadly classified as a spectrum of acute gastric mucosal injury. It is postulated that disruption of the mucosa and blood vessels within the submucosal layer results in dissection of the muscularis propria from the submucosa, with eventual clot formation. Progression of the haematoma was documented both endoscopically and surgically in an elderly anticoagulated patient who suffered a complication of therapeutic endoscopic intervention. A review of the literature is presented.

1. Introduction
Gastric intramural hematoma is an uncommon condition in current medical practice. Also termed intramural dissection, this dangerous condition may be more broadly classified as a spectrum of "acute gastric mucosal injury", where disruption of the mucosa and blood vessels within the submucosal layer results in dissection of the muscularis propria from the submucosa, with eventual clot formation. The condition has been reported throughout the gastrointestinal tract and can be broadly attributed to spontaneous, medical, traumatic or iatrogenic causes. While most cases resolve with conservative management, surgery was mandatory in a few described cases of gastric intramural haematoma due to concomitant perforation or gastric outlet obstruction. To the best of our knowledge, this is the second case documenting both endoscopy and surgery for uncontrolled haemorrhage.

2. Case report
An 81 year old Chinese female was admitted for a fall and managed by the orthopaedics unit at our institution for T12 and L2 compression fractures and a left clavicle fracture. Her prior medical conditions include hypertension, hyperlipidaemia, and osteoporosis. She was provided with analgesia including a course of NSAIDs. During this admission, she also developed a left segmental pulmonary embolus, in the absence of deep vein thrombosis, possibly related to immobility. After thorough investigations to rule out secondary causes, she was started on subcutaneous low-molecular-weight heparin (enoxaparin) at the dose of 1 mg/kg twice daily by her haematologist. Upon initiating treatment, she was found to have progressive iron deficiency anaemia hence an upper gastrointestinal (GI) endoscopy and CT colonography was arranged by her managing gastroenterologist. Enoxaparin was withheld for 12 h prior to the procedure.

Her upper GI endoscopy revealed pangastric erosions and a 4 mm ulcer with visible vessel in the gastric antrum. The Forrest 2a ulcer was treated with adrenaline injection and heater probe. Small biopsies were taken from fundus for urease testing and histology. Haemostasis appeared to be secure at the end of the procedure. An hour later, the patient developed tachycardia, haematamesis, and significant haemoglobin drop of 2 g/dL. An endoscopy was repeated immediately. This revealed large amount of blood clots in the gastric antrum which could not be completely removed by suctioning.

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Hence the source of bleeding could not clearly identified (Fig. 1a). She was actively resuscitated with fluids and transferred to the intensive care unit.

Angioembolisation was not entertained as a second line modality due to high risk of post-procedure ulcer perforation related to ischaemia. As there was no significant haemodynamic compromise after initial resuscitation with fluids, a second attempt at therapeutic intervention with upper GI endoscopy was performed under general anaesthesia in the operating room. This second upper GI endoscopy in the operating room again revealed a large amount of blood clots as well as on-going oozing of fresh blood. The endoscopist attempted to remove the clots with a snare to better localise the course of bleeding but was unsuccessful. A significant antral submucosal hematoma had begun developing along the anterior wall of the antrum (Fig. 1b). We were unable to localise the source of continuous fresh bleeding and decision was made for immediate exploratory laparotomy.

At laparotomy, it was apparent that a large haematoma had occupied the anterior aspect of the stomach causing extensive serosal stretch from fundus to pylorus (Fig. 2a). The lesser sac was entered and a gastrostomy was performed at the relatively normal posterior wall of the stomach. The tense intramural gastric haematoma was revealed within the stomach lumen. In addition, the mucosa of the anterior wall of the stomach was lacerated at three parts, demonstrating the tense blood clot spilling into stomach lumen. Small areas of active bleeding were controlled with direct pressure. Haemostasis was quickly achieved. A nasojejunal tube was inserted under direct visualization before closure of the gastrostomy.

The patient recovered well post-operatively. She was recommenced on prophylactic dose enoxaprin 48h after surgery. Proton pump inhibitors were prescribed for the management of her antral ulcer on the background of gastritis. There was no evidence of Helicobacter pylori colonisation. Nasojejunal feeding was continued for one week before diet was gradually escalated back to normal. She was discharged well 14 days later.

3. Discussion

First described by MacLauchlan in the Lancet in 1838 as a false aneurysmal tumour in the duodenum [1], a century later the condition is better understood. However, it remains difficult to obtain an accurate estimate of the incidence of intramural gastrointestinal haematomas as it persists to be a rare clinical and surgical entity. Postulated pathophysiology includes disruption of blood vessel within the submucosal layer which uncommonly results in dissection of the muscularis propria from the submucosa, resulting in a “false aneurysm”.

Intramural gastrointestinal haematomas have been described in the oesophagus, stomach, small bowel and colon. The most common site reported is the duodenum, with over 130 cases in the literature [2]. In these patients, blunt abdominal trauma was the most common causative association, and gastric outlet obstruction, intestinal obstruction and intussusception are known complications [2]. The second most common site is the oesophagus with over 79 cases [3], followed by isolated reports of 47 cases involving the stomach (Table 1).

In gastric intramural haematomas, a review of the literature suggests that the most common aetiology is coagulopathy, with or without contribution by peptic ulcer disease. This is most commonly related to use of anticoagulation [4–15], less commonly in patients with haemophilia [16–19], and one case described a patient with thrombocytopenia from myelofibrosis [20]. Other causes include peptic ulcer disease [21–23], vascular aneurysms [24–27], fish bone ingestion [28,29], as a complication of endoscopy [30–36], spontaneous and idiopathic cases [37–41], and other isolated cases related to amyloidosis [42–44], pancreatitis [45], Ehlers-Danlos syndrome [46], splenic rupture [47], and one case describing haematoma after splenectomy for idiopathic
thrombocytopenic purpura [48]. Ours is the first case reporting haematoma in a patient anticoagulated with enoxaparin, with contribution from peptic ulcer disease. We are also reminded that care should be taken during endoscopy in such patients to prevent potentially life-threatening complications. The condition should be clearly recognised to aid choice of management.

There is no agreed standard of care for this rare condition, however the majority of (64.5%) documented cases resolve spontaneously. Based on first principles, the managing clinician should first aim to resuscitate the patient and secure haemostasis. Meanwhile, medical management involves correcting any underlying coagulopathy. In the event of failure of conservative treatment, described therapeutic modalities to achieve haemostasis include endoscopy, arterial embolization, percutaneous drainage and surgery. From our analysis, most cases as a result of coagulopathy and fish bone ingestion were treated successfully with medical and supportive measures. Two cases out of 17 cases caused by coagulopathy were resolved by transarterial catheter embolization after bleeding was isolated to the left gastric arteries [8,12]. One case caused by focal pancreatitis benefited from percutaneous aspiration of the established haematoma [45]. Only one out of 7 cases caused by peptic ulcer disease and gastric aneurysms and one complication of endoscopy required laparotomy for perforation [22,34]. Most cases of amyloidosis resulted in gastrectomy for irretractable recurrent bleeding and outcomes in these patients were poor, including death due to multiorgan failure [42,43]. The patient with Ehlers-Danlos syndrome required surgery mainly because of gastric outlet obstruction from the haematoma, which also resulted in transient ischaemia of the duodenum due to increased intraluminal pressure that resolved upon clot evacuation [46].

Of particular interest to us are the idiopathic cases for which majority of patients were misdiagnosed to have gastric tumours. Because of diagnostic dilemmas, these patients likely received unnecessary gastrectomies due to lack of recognition of the condition and diagnoses were only revealed post-operatively [37–39].

In all cases, the decision to operate emergently was largely driven by the clinical picture and specific indications, such as gastric perforation, gastric outlet obstruction, and less commonly for unresolved bleeding. Our patient is such a rare case who benefited from surgical haemostasis. Following arrest of bleeding, one then selects an appropriate route of alternative alimentation while the patient undergoes bowel rest while awaiting resolution of the haematoma. This may comprise of either nasojejunal tube feeding or feeding jejunostomy.

4. Conclusion

Gastric submucosal haematoma is a rare disorder that should be recognised to avoid unnecessary surgical intervention and misadventure, as most cases may be managed conservatively.

Conflict of interest

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Consent

Consent has been obtained.

Author contribution

First author – writing main bulk of the paper. Second author – study concept and design, writing the paper. Corresponding author – editing and making necessary changes to the paper.

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