**Manuscript:**

**Abstract:**

Splenic artery aneurysm and pseudoaneurysm are rare pathologies and uncommon causes of massive upper gastrointestinal bleeding. They represent the third most common intra-abdominal aneurysms. Variations in the origin of splenic artery are relatively common and asymptomatic. However, the presence of an accessory splenic artery that is symptomatic is quite atypical. In this report, we describe the case of a 73-year-old female who presented with massive upper gastrointestinal bleeding caused by a pseudoaneurysm of a superior polar artery with an unusual anatomical origin. The patient was successfully treated endovascularly with transarterial coil embolization. Early diagnosis and interventional management is crucial for patient’s survival; hence, it must be kept in mind as possible etiology of life-threatening gastrointestinal bleeding to reduce morbidity and mortality.

**Key words:** Superior polar artery, pseudoaneurysm, GI bleeding, embolization

**Introduction:**

Acute upper gastrointestinal (GI) bleeding is a potentially life-threatening emergency that remains a common cause of hospitalization—most cases being attributed to peptic ulcer disease or gastric/esophageal varices. Although rare, aneurysm of the splenic artery (SA) has been described as a cause of acute upper GI hemorrhage that, when diagnosed, represents a potential fatal event given the risk of massive bleeding due to rupture [1]. SA aneurysm is the most common intra-abdominal splanchnic aneurysm and the third most common intra-abdominal aneurysm following those involving the aorta and iliac vessels [1,2]. Variations in the origin of SA are common. However, anatomical variation in its branching pattern is relatively rare [3]. Knowledge of these anatomical variations to successfully approach surgical/interventional planning and awareness of this entity as a potential GI bleeding etiology is crucial.

The SA is the largest branch of the celiac trunk that courses along the superior border of the pancreas. Near the splenic hilum, it gives 2-3 terminal branches that further divides into 4-6 segmental intra-splenic branches [4,5]. Rare accessory arteries have, however, been documented.

A superior polar artery (SPA) as an anatomical vascular variant usually arises from the main SA. It generally arises approximately 2-3 cm from the splenic hilum with an average length of 3.9 cm and supplies the upper pole of the spleen directly without passing through the hilum [5].

Similar to the case described by Baidwan et al. [5], in this report we present an uncommon variant of the SPA that measured approximately 8 cm long, originating at approximately 6 cm from the splenic hilum. More interestingly, it presented with a pseudoaneurysm that ruptured causing massive upper GI bleeding.

Few cases of upper GI bleeding related to ruptured accessory SA aneurysms have been described [4]. To our knowledge, this would be the third report in the medical literature describing massive upper GI bleeding caused by a pseudoaneurysm of an accessory SA to the upper pole of the spleen; the first arising from a superior polar artery successfully treated with transarterial coil embolization.

**Case Description:**

A 73-year-old female with no history of cirrhosis, but a positive history of ischemic cardiomyopathy, congestive heart failure, type 2 diabetes mellitus, and atrial fibrillation, on warfarin (Coumadin, 3.5 mg daily) was admitted to our institution for nausea, shortness of breath, chest pain, and hematemesis. No melena, fever, or lower extremity edema was present. The patient reported vomiting up large amounts of dark red blood, with clots, 3 hours earlier resulting in her calling an ambulance. While in transit, she had another episode in which she vomited an additional liter of dark red blood. Upon arrival at the emergency department (ED) at 7:16 PM (day 1), she was found to have weak peripheral pulses, tachycardia (123 bpm), and hypotension (82/54 mmHg). Physical examination revealed mild epigastric tenderness with a soft, non-distended abdomen and hyperactive bowel sounds.

Laboratory data demonstrated mild anemia with a hemoglobin (9.3 g/DL) and hematocrit (27.2%), as well as slightly elevated prothrombin time (20.6 sec) and International Normalized Ratio (1.8). White blood cell count (7.4 x 109/L) and platelet count (226 x 109/L) were within normal range. CK-MB (6.6 ng/mL) and troponin (1.630 ng/mL) were elevated.

In spite of additional transfusion with two units of p-RBCs, the patient’s hemoglobin remained mildly low (9.3g/dL). There continued to be intermittent bloody output through NG tube. The patient was therefore referred to Interventional Radiology for emergent visceral angiography with intervention.

**Procedures:**

**Esophagogastroduodenoscopy:** In the ED, the patient was given intravenous boluses of a large volume of fluid (3L of normal saline) and transfused with two units of packed red blood cells (p-RBCs). Following nasogastric (NG) tube placement, the patient underwent esophagogastroduodenoscopy (EGD) the following morning (day 2) at 10:47 AM, which demonstrated a small vessel in the distal esophagus with stigmata of recent bleeding. It was injected with 2 mL of epinephrine (1:10,000) and clipped. Although a small amount of thrombus was found in the gastric fundus, there was no source of bleeding identified on EGD. Additionally, the duodenum was normal. Towards the end of the endoscopic evaluation the patient became increasingly tachycardic (135 bpm) and hypotensive (systolic blood pressure < 65 mmHg). The procedure was terminated and the patient was admitted to the cardiac intensive care unit.

**Visceral Angiography with Embolization (11:44 AM):** Under moderate sedation and with local anesthesia, right femoral arterial access was obtained percutaneously. Digital subtraction angiography (DSA) of the descending aorta was performed using a 5 Fr pigtail catheter (Soft-Vu Omni Flush, AngioDynamics, Latham, NY); it demonstrated a small pseudoaneurysm (6 mm) at the gastric fundus arising from a superior polar artery originating from the proximal segment of the main SA at 6 to 7 cm from the splenic hilum and approximately 8 cm long, coursing toward the gastric fundus (**Fig. 1A**). The celiac trunk was catheterized with a 5-French reverse curve catheter (SIM-1, Cordis, Miami Lakes, FL). Intragastric contrast extravasation from the pseudoaneurysm was identified in the distal segment of this artery and demonstrated curvilinear appearance as it pooled in the gastric rugae, mimicking the appearance of a vein (**Fig. 1B**). A 2.4-French coaxial microcatheter system (Progreat, Terumo, Tokyo, Japan) was used to catheterize the superior polar branch of the SA, and a microcoil (Nester Embolization Coil, Cook Medical, Bloomington, IN) was deployed. A final angiogram demonstrated complete occlusion of the target artery with no opacification of pseudoaneurysm or extravasation of contrast (**Fig. 1C**). Total procedure time was 1 hour and 30 minutes.

Over the course of several days, following the procedure, the patient’s condition improved and her hemoglobin stabilized without further transfusion. The patient was discharged 7 days after the procedure and no recurrent symptoms were reported at 4-month follow-up.

**Discussion:**

GI bleeding due to rupture of a SA aneurysm is a very uncommon, potentially fatal vascular complication that poses diagnostic and therapeutic challenges. A quick diagnosis and treatment is imperative because of its association with high morbidity and mortality rates [6].

SA aneurysms are the third most common intra-abdominal aneurysm behind aneurysm of the aorta and iliac arteries. The majority (60%) are solitary and < 2 cm in diameter and 75% are found in the distal third of the SA incidentally. Twenty percent (20%) are located in the middle third [7].

The SA represents the largest branch of the celiac trunk, running along the superior aspect of the pancreas before reaching the splenic hilum through the splenorenal ligament. Studies on the origin variation of the SA are multiple. However, anatomical variations in its branching pattern are very rare and even rarer are symptomatic manifestations of such variants [4]. A study on this subject reported that, in the majority of cases, it arises from the celiac trunk (90.6%). Rarely, it has origin from the abdominal aorta (8.1%) and from other sites such as the common hepatic artery, left gastric artery, or superior mesenteric artery [8,9]. In a few cases, the spleen is supplied by accessory splenic arteries with cases reported originating from the left gastroepiploic artery, left gastric artery, or polar arteries arising primarily from the SA [4,10,11].

Most of the SA aneurysms arise from the distal and middle third of the SA. Of the few rare cases of accessory SA variations, only two cases of an aneurysm from an accessory SA arising from the left gastric artery presenting as massive upper GI bleeding have been reported [4,11]. However, none has been documented arising from a superior polar artery of the spleen.

There is controversy regarding the description of the origin of the polar artery and its nomenclature. A superior polar artery from the SA usually arises from the distal splenic artery, near the hilum, but it may originate from the superior terminal branch of the SA. It is quite constant, generally arising 3-5 cm from the splenic hilum with average length of 3.9 cm. It supplies the upper pole of the spleen directly without passing through the hilum [5].

The incidence of the presence of the superior polar artery is variable, ranging between 31.3% and 65% [5]. In the study conducted by Sahni et al. [9], In 102 of 200 (51%) specimens, the superior polar arose from the SA just distal to the origin of the posterior gastric artery and about 4-5 cm proximal to the hilum of the spleen. In 3 of 200 (1.5%) specimens, it arose as a branch of the posterior gastric artery. Others have stated that terminal branches from the SA anywhere within 1-12 cm can be regarded as a polar artery [10]. Regardless the diverse nomenclature, the fact remains these branches divide the spleen into definite vascular segments. Detailed knowledge of these segments is useful for segmental resection, as well as the now-favored segmental arterial embolization of the spleen, which preserves splenic tissue as much as possible [3].

Baidwan et al. described in their cadaveric specimen a rare variation of the superior polar artery which was 7.6 cm long and 6.4 cm proximal to the splenic hilum [5]. In our case, with the use of DSA, we estimated that the polar artery measured approximately 8 cm long and was located 6 to 7 cm from the splenic hilum. This uncommon variation of the superior polar artery also presented as a pseudoaneurysm that had ruptured provoking massive upper GI bleeding. Although EGD could not localize an active bleeding source, it is possible that a small and inconspicuous penetrating ulcer in the gastric wall caused an injury to the superior polar artery, which had an intragastric course through the wall of the gastric fundus, provoking intraluminal hemorrhage. To our knowledge, this case is the third report in the medical literature showing an accessory SA to the upper pole of the spleen presenting with massive upper GI bleeding due to a ruptured pseudoaneurysm and the first from a superior polar artery.

CTA is the best modality for evaluation of the splanchnic vascular anatomy and diagnosis of un-ruptured aneurysms. It provides valuable information regarding the diagnosis of this pathology and localization for surgical or vascular intervention. Magnetic resonance angiography is highly sensitive and specific. Nevertheless, limitations such as a long study time and availability make it less feasible, especially in emergency patients. Multi-detector CT provides excellent image quality and volume-rendered image reconstruction, which is advantageous for the diagnosis. CT angiography has been reported to have a sensitivity of up to 94.7% and a specificity of 90.0% [7].

**CONCLUSION**

The most common causes of upper GI bleeding in the ED are related to either gastric/esophageal varices or gastric peptic ulcer disease. In patients presenting with upper GI bleeding, recognition of aneurysms or pseudoaneurysms originating from the SA and its anatomical branching pattern should now be considered as possible etiology. It is likely that these lesions can have a relapsing course, which may render them undetectable on endoscopy and undetected if asymptomatic. CTA is the primary modality for evaluation of variant anatomy of splenic and accessory splenic arteries and diagnosis. Knowledge of these arteries becomes clinically important to avoid dangerous and potentially fatal bleeding when performing surgical and radiological procedures.

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**Figure Legends:**

**Figure 1. A** Digital subtraction arteriogram (DSA) of a 73-year-old female demonstrates a 6-mm pseudoaneurysm (short arrow) of the superior polar splenic artery (long arrow) that is arising from the proximal part of the main splenic artery (double arrows). **B.** On delayed arteriogram, extravasation of contrast material from the pseudoaneurysm (short arrow) demonstrates a tubular appearance with varying widths as it pools in the gastric rugae (long arrows), comparing to more straight and smooth appearance of the splenic vein (double arrows). **C.** Follow-up DSA after transarterial embolization demonstrates complete occlusion of the superior polar splenic artery with a microcoil placed across the neck of the pseudoaneurysm (arrow).