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Case Report

Acute upper gastrointestinal hemorrhage from a pseudoaneurysm of an unusual superior polar artery of the spleen

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ABSTRACT

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

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 and 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, anatomic variation in its branching pattern is relatively rare [3]. Knowledge of these anatomic variations to successfully approach surgical or 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 intrasplenic branches [4,5]. Rare accessory arteries have, however, been documented.

A superior polar artery (SPA) as an anatomic vascular variant usually arises from the main SA. It generally arises

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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, which 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 an SPA successfully treated with transarterial coil embolization.

Case description

A 73-year-old female with no history of cirrhosis, but with 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 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, nondistended 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 seconds) and international normalized ratio (1.8). White blood cell count (7.4×10^9/L) and platelet count (226×10^9/L) were within normal range. Creatine kinase-muscle/brain (CK-MB) (6.6 ng/mL) and troponin (1.630 ng/mL) were elevated.

In spite of additional transfusion with 2 units of packed red blood cells, the patient's hemoglobin remained mildly low (9.3 g/dL). Intermittent bloody output through the nasogastric tube was noted. 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 (3 L of normal saline) and transfused with 2 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. Toward 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 of the descending aorta was performed using a 5-Fr pigtail catheter (Soft-Vu Omni Flush, AngioDynamics, Latham, New York); it demonstrated a small pseudoaneurysm (6 mm) at the gastric fundus arising from an SPA originating from the proximal segment of the main SA at 6-7 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-Fr reverse-curve catheter (SIM-1, Cordis, Miami Lakes, Florida). 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-Fr 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, Indiana) 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%) is solitary and <2 cm in diameter, and 75% are found in the distal third of the SA incidentally. Twenty percent 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.

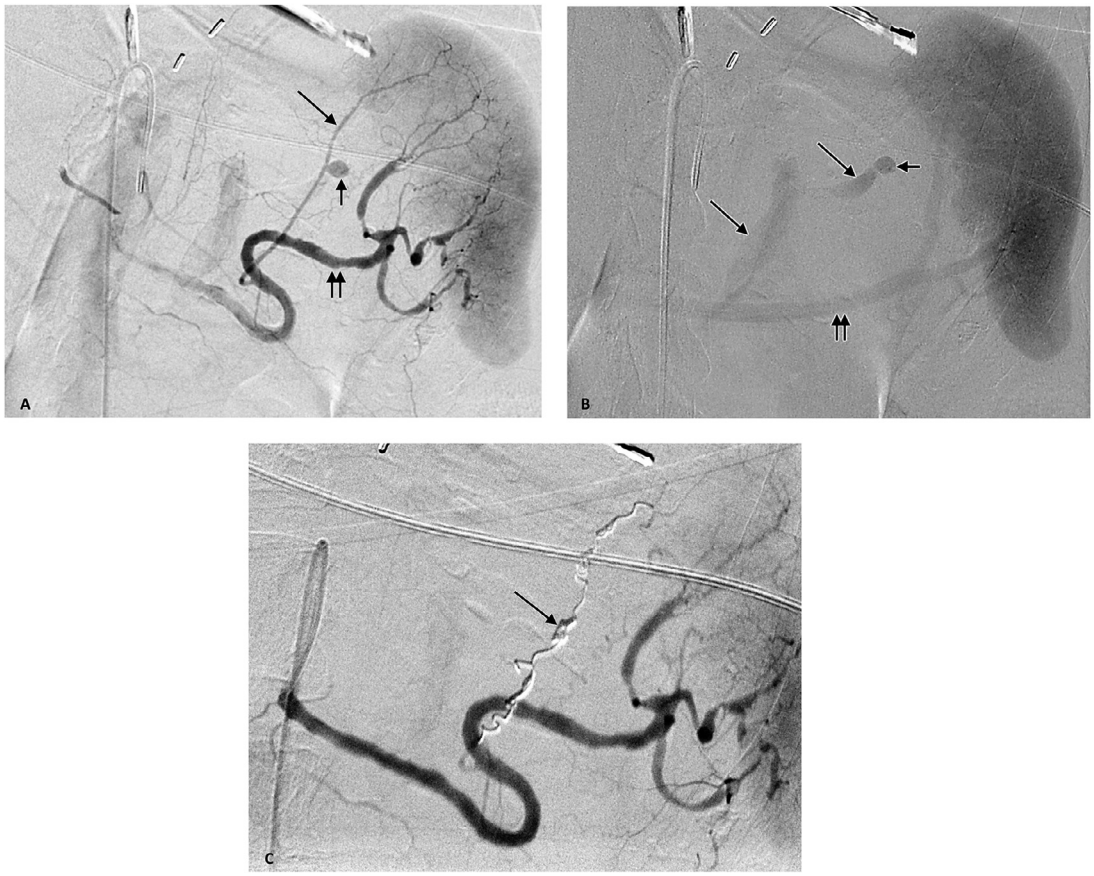

Fig. 1 – (A) Digital subtraction arteriogram of a 73-year-old female demonstrates a 6-mm pseudoaneurysm (short arrow) of the superior polar artery (SPA; long arrow) that is arising from the proximal part of the main SA (double arrows). (B) On delayed arteriogram, extravasation of contrast material from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

However, anatomic variations in its branching pattern are very rare, even symptomatic manifestations of such variants are rarer [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 SAs 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 2 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 an SPA of the spleen.

There is controversy regarding the description of the origin of the polar artery and its nomenclature. An SPA from the SA usually arises from the distal SA, 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 SPA is variable, ranging between 31.3% and 65% [5]. In the study conducted by Sahni et al. [9], in 102 of 200 specimens (51%), 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 specimens (1.5%), 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 SPA, which was 7.6-cm long and 6.4-cm proximal to the splenic hilum [5]. In our case, with the use of digital subtraction angiography, we estimated that the polar artery measured approximately 8-cm long and was located at 6-7 cm from the splenic hilum. This uncommon variation of the SPA

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 SPA, 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 an SPA.

CTA is the best modality for evaluation of the splanchnic vascular anatomy and diagnosis of unruptured 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. Multidetector computed tomography provides excellent image quality and volume-rendered image reconstruction, which is advantageous for the diagnosis. Computed tomography 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 and 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 anatomic 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. Computed tomography angiography (CTA) is the primary modality for evaluation of variant anatomy of splenic and accessory SAs 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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