

Abstract

The inferior phrenic artery (IPA) aneurysms are rare visceral aneurysms. It usually occurs as a secondary to trauma, surgery or as a complication of pancreatitis, systemic pathology such as vasculitis, collagen vascular disorders, sepsis or segmental arterial mediolysis (SAM). SAM is a non-inflammatory vascular injury secondary to repeated vasospasm and is related to hypertension. IPA presents with abdominal pain, melena, hematochezia, anemia. The ruptured and actively bleeding aneurysm can lead to hemorrhagic shock and immediate management is required with angiography and endovascular embolization with coil or gel foam or stent. Inaccessible locations are reached with surgical intervention, but it is associated with high mortality and morbidity.

Introduction

Visceral artery aneurysms and pseudoaneurysms are uncommon but potentially lethal clinical entities.

IPA pseudoaneurysm can be due to:

- catheter-based interventions
- complication of pancreatitis[1]
- trauma
- surgeries like coarctectomy[4,5] or gastrectomy[1,6].
- sepsis
- vasculitis (Polyarteritis nodosa, Takayasu's arteritis, Behcet's syndrome, and Henoch-Schoenlein purpura) [3]
- Collagen vascular diseases
- Segmental Arterial Mediolytic (SAM)

Visceral artery aneurysm can be present along the celiac artery and its branches (39%), hepatic (39%), splenic (18%) and superior mesenteric artery (4%), celiac trunk (4%), gastric and gastropiploic arteries (4%), intestinal arteries (3%), pancreaticoduodenal arteries (2%), gastroduodenal artery (1.5%), and the inferior mesenteric artery (1%) [2].

Case Presentation

50-year-old male presented with one-day history of epigastric and left upper quadrant abdominal pain. Review of symptoms positive for unintentional weight loss of 20lbs in the past six weeks. Medical history is significant for hypertension, untreated Hepatitis C infection, intravenous drug abuse, and smoking. No history of pancreatitis, blunt trauma, abdominal surgery, and alcohol use. On examination, his vitals were stable, diminished left lung base breath sounds and epigastric and LUQ abdominal tenderness without peritoneal signs. Laboratory workup showed elevated WBC count, creatinine 1.15 mg/dl, mildly elevated ALT and AST, positive Hepatitis C antibody and normal lipase level and coagulation panel. The urine drug screen was positive for methamphetamine and opioids. CT with contrast (Fig 2) showed active bleeding at gastric cardia with posterior mediastinal and distal esophagus/gastric cardia hypodensity concerning for hematoma. Small volume, high-density fluid along the greater curvature of the stomach compatible with hemoperitoneum(Fig 3). He underwent esophagogastroduodenoscopy revealing no active bleed. Subsequently, he underwent a celiac, left gastric, splenic, multilevel Intercostal, and IPA angiogram. An aneurysm of the left IPA with contrast extravasation was identified, and embolized using coil and gel foam(Fig 1).

Radiological Findings



Fig 1. Angiography of left phrenic artery shows aneurysm and extravasation of contrast indicating hemorrhage

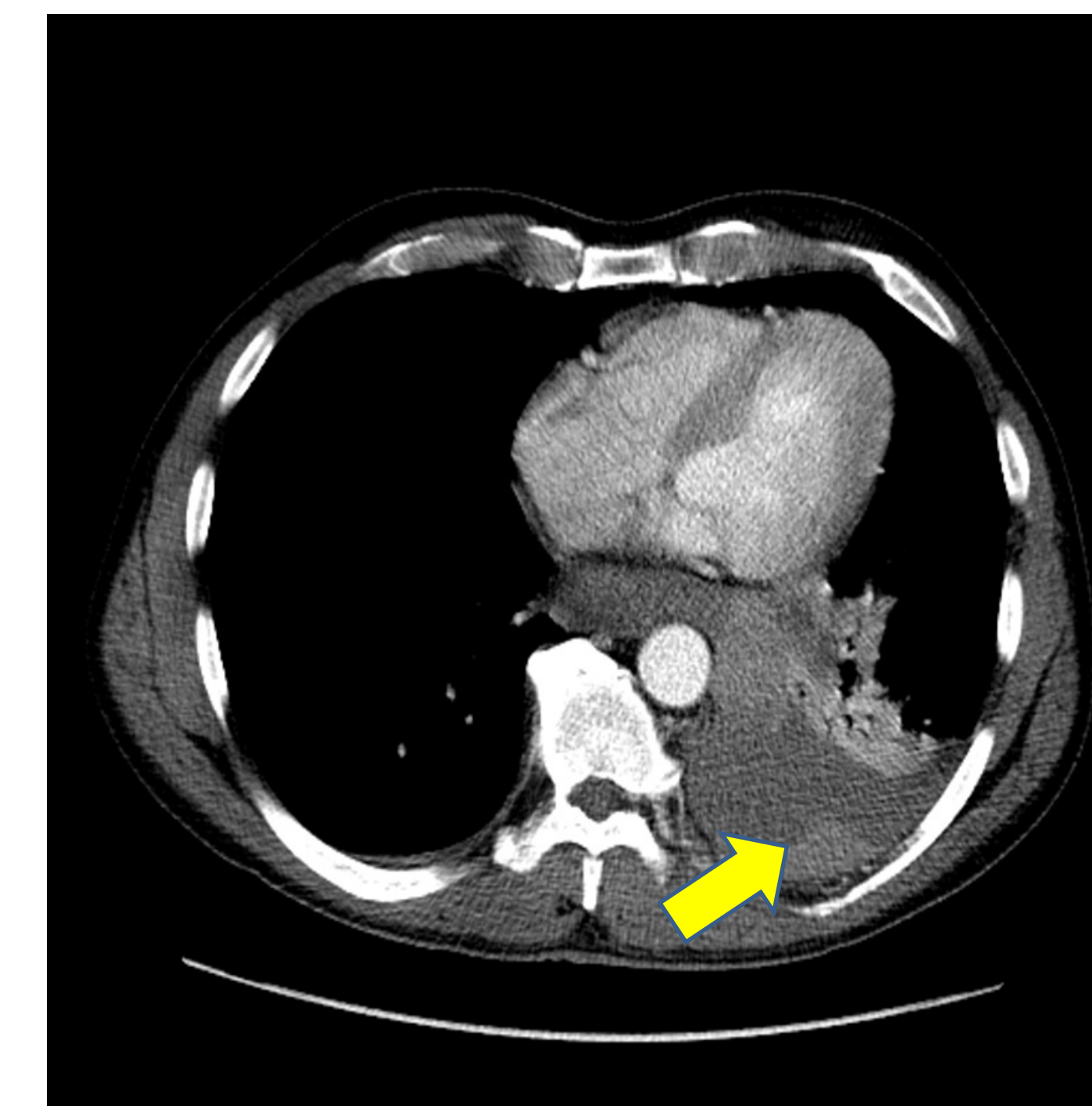


Fig 2. CT chest with contrast showing moderate sized complex left hemithorax with layering high density



Fig 3. CT Abdomen and pelvis with contrast showing posterior mediastinal mass extending into abdomen yellow arrow

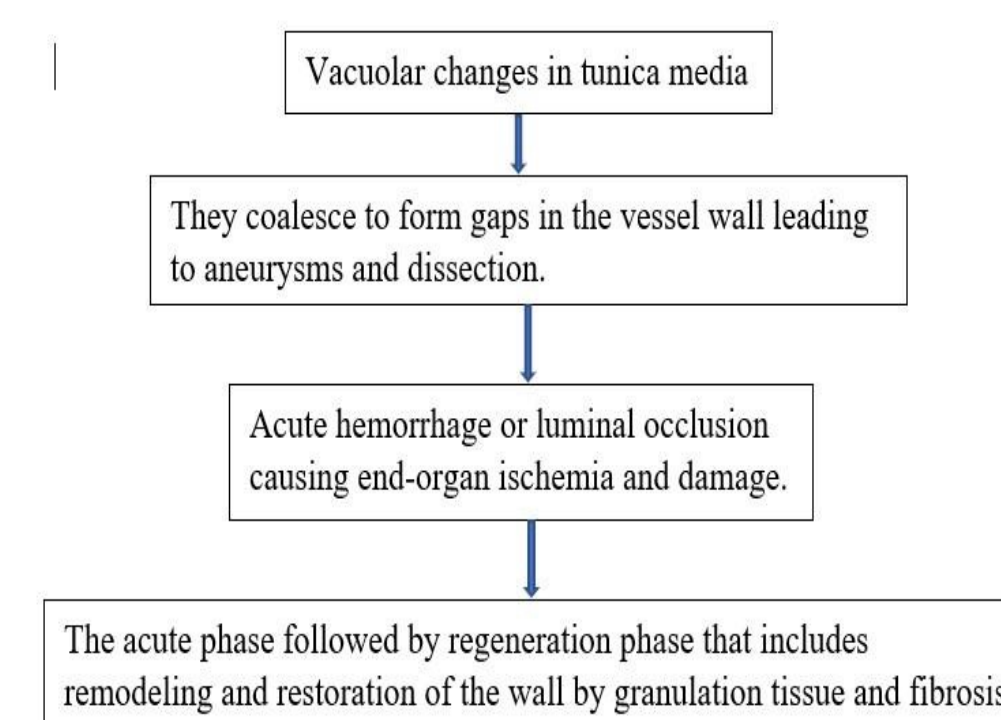


Fig:4 Pathophysiology of Segmental arterial myelinolysis.

Discussion

Segmental Arterial Mediolytic is an idiopathic noninflammatory, nonatherosclerotic vascular disease. Hypertension is noted in 43% of cases diagnosed with SAM [7]. The pathophysiology is described in Fig 4. The splanchnic arteries are commonly involved in SAM [8]. Our patient has no prior history of trauma, surgeries, or pancreatitis. Vasculitis at can lead to aneurysm pseudoaneurysm formation [9]. Vasculitis is excluded with normal inflammatory markers and the absence of other symptoms. Ehler-Danlos syndrome is due to abnormal collagen formation (COL5A genes), causing defective vessel formation and aneurysms. However, splanchnic vasculature involvement is very rare [10], and our patient has no suggestive clinical features or family history. Ruling out all the alternate diagnoses we suspect the etiology of aneurysm could be due to SAM, given the history of hypertension.

The rupture of an aneurysm may occur spontaneously or after exertion. Contrast enhanced angiography is the first line of investigation. Catheter angiography remains the gold standard in evaluating and as a therapeutic modality for aneurysms [11].

Embolization is done in cases with active bleeding [12]. Surgical intervention has a higher rate of postoperative morbidity and mortality [12].

Conclusions

Inferior phrenic artery aneurysm can be due to various causes. It could present as mass compressing surrounding structures or hemorrhagic shock or as a well contained hematoma. The diagnosis is based on the patient's history, clinical examination and laboratory findings. Irrespective of etiology, the initial step is imaging and emergency management with angiography. The endovascular intervention using coils, or gel foam plays a vital role and decreases the mortality rate associated with ruptured aneurysm. The specific diagnosis is by histopathological examination after resection or postmortem.

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