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Primary Aorto-enteric Fistula due to a Mycotic Infrarenal Aortic Aneurysm: A Rare Surgical Nightmare

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Abstract

Background: Primary aorto-enteric fistula is a rare condition, with fatal outcomes without surgical management. *Case Report*: We present one such case where an elderly patient presented with pain abdomen and a 'herald bleed', with a mycotic aortic aneurysm, warranting emergency surgery. *Conclusion*: Emergency surgical management is the only interventional modality that plays a pivotal role in preventing mortality.

Keywords: Aortic Aneurysm, Mortality, Mycotic Aneurysm, Pain, Vascular Surgery.

Introduction

This report describes the management of a primary aorto-enteric fistula (PAEF), a rare but life-threatening pathology, defined as a communication between the aorta and gastrointestinal tract. First described in 1829, the reported incidence is 0.04-0.07%, with approximately 350 reported cases [1]. Proximity with the enteric system makes the fistulous communication common with third and the fourth part of duodenum, followed by jejunum and ileum. Clinically, aorto-enteric fistula presents with triad of symptoms, first explained by Sir Astley Cooper, however a 'herald gastrointestinal bleed' can be catastrophic [2]. In view of rarity of the entity, management protocols are based on case reports and case series. In this case we revascularized the lower limbs with an extra-anatomical bypass, in view of a soiled retroperitoneum, and resected the mycotic aortic aneurysm [3].

Case Report

A 62-year-old gentleman presented with complaints of diffuse pain abdomen, low grade fever and vomiting of one week duration. He had one episode of bleeding per rectum one day prior to his hospital visit. He also gave history of and unquantifiable weight loss and anorexia of one week duration. He was being evaluated for his fever and pain abdomen when the episode of bleeding per rectum occurred and in anticipation of it being a 'herald bleed' his treating clinician ordered a computed tomography angiography (CTA) which revealed an infrarenal saccular abdominal aortic aneurysm with loss of fat planes, with abutting fourth part of duodenum. He was hence referred to this vascular surgery centre for emergency management. On examination he had diffuse abdominal pain, with no guarding or rigidity. He had tachycardia and hypotension. On palpation of abdomen a pulsatile peri-umbilical swelling (8×6 cm) was palpable. All peripheral pulses were palpable.

His haematological reports revealed leucocytosis (total leucocyte count: 18.12×10^3/L) and anemia (hemoglobin: 7 mmol/L). Other markers of inflammation like erythrocyte sedimentation rate

and serum procalcitonin were also raised. A CTA revealed circumscribed thickening of abdominal aorta (93 mm long segment) with fusiform dilatation (52×45 mm) caudal to aortic bifurcation with a saccular dilatation of infra-renal abdominal aorta (20×18×22 mm) with peripheral thrombus and fat stranding. There was tethering of adjacent small bowel loops towards the aneurysm with loss of fat planes with duodenum. A right renal calculus (7×14 mm) was additional finding **[Fig.1]**. 2-D echocardiography revealed grade one diastolic dysfunction, with mild tricuspid regurgitation.

Following discussion with a multi-disciplinary team of gastrointestinal surgeon, intensivists, and cardiologist an emergency extra-anatomical revascularization of lower limbs with an axillo-bifemoral graft, resection of mycotic abdominal aortic aneurysm, retroperitoneal debridement and closure of bowel defect was adjudged as the ideal treatment, in prevailing clinical and hemodynamic status of the patient. After volume resuscitation and under broad-spectrum antibiotic cover the preliminary extra-anatomical bypass was provided by an extended polytetrafluoroethylene (ePTFE) (8×55 mm) inter-ring bypass graft by an experienced vascular surgery team **[Fig.2]**. Through a midline laparotomy early proximal aortic control was obtained through an infra-renal clamping. Intra-operatively, the mycotic aneurysm was found extending from 1.5 cm distal to renal arteries to 1 cm proximal to aortic bifurcation. Dense peri-aortic adhesions and a primary aorto-enteric fistula between the third part of duodenum and aneurysmal sac was delineated. The mycotic infra-renal abdominal aortic aneurysm (IRAAA) was resected. After mycotic aneurysmal sac was excised, and retroperitoneal debridement was done the proximal and distal aortic stump were closed. As the duodenal defect was small it was closed primarily and omental interposition between the aortic stump and enteric contents was done **[Fig.3]**. The abdomen was closed over drains. As planned

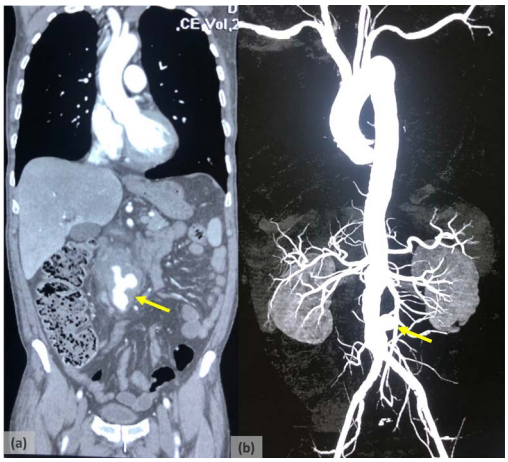

Fig.1: *Computed tomography images of the PAEF in coronal section.* **(a):** *the infrarenal aortic aneurysm with effacement of fat planes with duodenum and soft tissue thickening (marked with yellow arrow);* **(b):** *reconstructed aortogram images showing the saccular infrarenal aortic aneurysm (marked in yellow arrow).*

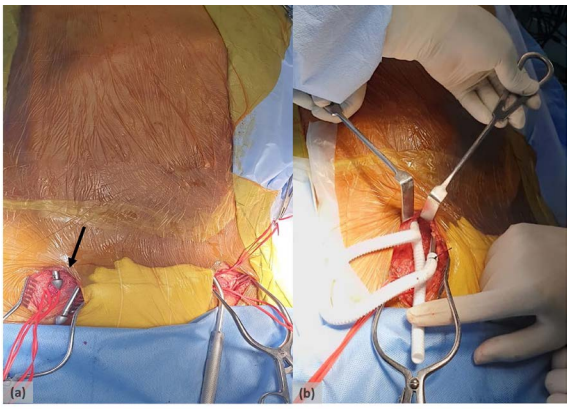

Fig.2: *Intra-operative pictures of extra-anatomical revascularization in the form of axillobifemoral bypass* **(a):** *the subcutaneous tunnels constructed with tunnelers (marked by black arrows);* **(b):** *the ePTFE graft being tunneled.*

the patient was shifted to intensive care unit following surgery on mechanical ventilatory and high inotropic support.

Patient had a stormy post-operative period and required high ionotropic support. He was managed in consultation with cardiac care team for a post-operative cardiogenic shock. The patient recuperated well and was discharged from the hospital, with preserved lower limb vascularity and a healed abdominal wound.

Discussion

First described by Sir Astley Cooper as a 'sometimes but serious complication of an aneurysmal aorta', in 1829, PAEF has always offered unmatched challenges to vascular surgeons [2]. The reported incidence of PAEF is 0.04% - 0.07% in large autopsy series and there is a limited availability of literature on the subject. Tumors, radiotherapy, and infection have been reported as the etiological factors [3,4]. The tethering effect of ligament of Treitz makes the third and fourth part of duodenum exposed to the pulsatile effects of aorta and eventually the commonest site for development of fistula (54%) followed by esophagus (28%) and stomach (2%). Despite the proposed mechanisms of mechanical, infectious, and inflammatory etiologies, the exact pathogenesis of PAEF is uncertain. In this case the infectious pathology of mycotic aneurysmal dilatation of the abdominal aorta was the inciting mechanism for the PAEF. Sir Astley Cooper described a classical clinical triad for PAEF which includes gastrointestinal bleeding (60-90%), abdominal pain (30-50%) and pulsatile abdominal mass (17-25%), however this triad is only seen in 11% patients [3,4]. Our case had two components of the triad in the form of gastrointestinal bleed and abdominal pain. Other associated symptoms comprise of back pain, fever, weight loss, easy fatiguability and sepsis [5].

CTA with a 60% detection rate, highest of all investigative modalities, assists in revascularization planning. The findings suggestive of effacement of aortic fat planes with soft tissue thickening and tethering of adjacent bowel loops, helps in clinching the diagnosis. Extravasation of contrast from the aorta is rare [3,6]. In view of hemodynamic instability of our patient the other modalities like esophagogastroduodenoscopy could not be performed. Data on laboratory findings is even more meagre, with hemoglobin below 8 mmol/L and leucocytosis reported in two-thirds and one-quarter of the patients of PAEF [3]. Our case had leucocytosis and anemia due to the ongoing sepsis and blood loss in gastrointestinal tract.

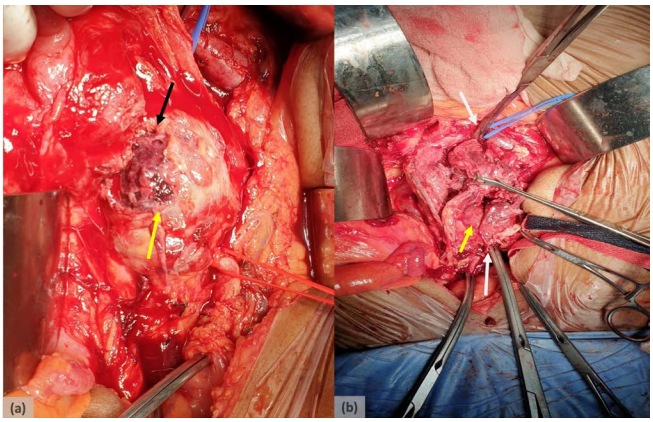

Fig.3 (a): *The mycotic infrarenal aortic aneurysm (marked with yellow arrow) with the PAEF (marked with black arrow);* **(b):** *the mycotic aneurysm after being laid open with the proximal and distal aortic clamps (marked by white arrow).*

The golden management goal ascribed to PAEF is 'to preserve life and limb salvage should be secondary'. A high index of clinical suspicion usually clinches the diagnosis. Surgery is the only interventional modality that has the probability of reducing fatal outcomes. Despite their being little consensus in the contemporary management of PAEF, timely surgical intervention plays a pivotal role in defining the outcome [3]. Multiple management strategies are in vogue as there are many schools of thoughts [7]. In view of gross retroperitoneal soiling, aneurysm resection, retroperitoneal toileting, omentoplasty and an extra-anatomical revascularization has been recommended and the same was offered to the patient by the authors. Endovascular repair is mostly reserved as a bridge to open surgical repair after initial resuscitation and infection control of hemodynamically stable patients [8].

Conclusion

PAEF is a rare clinical condition with certain mortality without surgical intervention. Timely diagnosis and surgical intervention are imperative. CTA with faster scans and ubiquitous presence forms the diagnostic modality of choice. The operative

dictum: save life first, limb later, formulates the guiding surgical principle. A situation tailored surgical management with multi-disciplinary team approach is necessary for a suitable outcome.

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