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Case Report

Celiac Arterioenteric Fistula after Open Repair of Celiac Artery Aneurysm – Case Report

Abstract

In a patient with a known history of aortic surgery, presence of upper gastrointestinal bleeding requires a high index suspicion for the possibility of aorto-enteric fistula. Aorto-enteric fistula is an uncommon but known complication occurring after abdominal aortic reconstruction. However, there are few reported cases of enteric fistulas arising after splanchnic artery aneurysm repair.

We report a unique case of a celiac artery graft – duodenal fistula in a 60-year-old male, who developed upper gastrointestinal bleeding two months after initial open resection of a celiac artery aneurysm and placement of an aorto-celiac artery graft. The patient underwent successful repair of the fistula and the resection of the involved graft with ligation of the common hepatic artery and splenectomy.

Our case report highlights the rare entity of celiac arterio-enteric fistula after open repair of a celiac artery aneurysm and reviews the diagnostic and treatment modalities available for successful management of this rare complication.

Introduction

Celiac artery aneurysm was first described by Bergeon in autopsy specimens in 1830 [1]. Celiac artery aneurysms are the 4th most common type of splanchnic artery aneurysms [2]. Celiac artery aneurysms account for 4-6% of splanchnic artery aneurysms [2,3]. The prevalence of celiac artery aneurysm is 0.005 to 0.05% [3].

In the first half of the 20th century, infection (syphilis) was the major cause of celiac artery aneurysms [2,4]. During this period, majority of patients presented with symptoms secondary to rupture of celiac artery aneurysm. Advancements in diagnostic modalities and treatment options during the second half of the 20th century has significantly changed the presentation and outcome of patients with celiac artery aneurysms. Currently, atherosclerosis and medial degeneration account for the majority of celiac artery aneurysms [1-4]. With the advent of computed tomography, majority of celiac artery aneurysms are detected incidentally before they have ruptured.

In 1958, Shumacker described the first successful surgical treatment of celiac artery aneurysm [3]. Surgical intervention in the form of ligation with or without revascularization with prosthetic graft is currently indicated for symptomatic aneurysms, aneurysms increasing in size, or aneurysms greater than 3 to 4 times the normal diameter of the celiac artery (8 mm) [3,4]. Endovascular treatment of celiac artery aneurysm with endovascular stent graft or coil embolization is an acceptable alternative in high-risk surgical patients [5].

Extensive experience with aortic aneurysm and its surgical repair has brought attention to the entity of aortoenteric fistula. While there are many cases of secondary aortoenteric fistulas reported in the literature, an extensive search of the literature with the terms "splanchnic artery aneurysm fistula", "celiac artery aneurysm fistula", "celiac artery aneurysm" and "aortoenteric fistula" revealed no case reports describing secondary arterioenteric fistulas arising after open celiac artery aneurysm repair. The extremely low incidence of celiac artery aneurysm and similarly low incidence of secondary fistula after repair of aneurysms may account for the paucity of literature on this topic. We describe a unique case of a secondary celiac arterioenteric fistula below.

Case Presentation

A 60 year old male underwent open resection of celiac artery aneurysm with placement of prosthetic graft between the aorta and the junction of common hepatic artery and splenic artery (Figures 1a,1b). Two months after the surgery, the patient presented to our institution with 2 week history of abdominal pain, fever, and unintentional weight loss. His past medical and surgical history was significant for stent placement for bilateral common iliac artery aneurysms. Computed tomography of the abdomen revealed a fluid collection with gas bubbles in the lesser sac adjacent to the graft site raising suspicion for graft infection (Figures 2a,2b,2c).

Within twelve hours of admission, the patient developed two episodes of hematemesis. The patient remained hemodynamically stable. Physical exam revealed no peritoneal signs. The patient's hemoglobin and hematocrit were stable at 9.4 g/dL and 29.6% respectively. The patient was emergently taken to the operating room.

Intra operatively, a selective celiac artery angiogram was performed and showed a ring like stenosis at the distal graft anastomosis. There was no contrast extravasation noted. Intra-abdominal exploration revealed two discrete foul-smelling fluid collections containing fresh blood in the lesser sac. Further evaluation

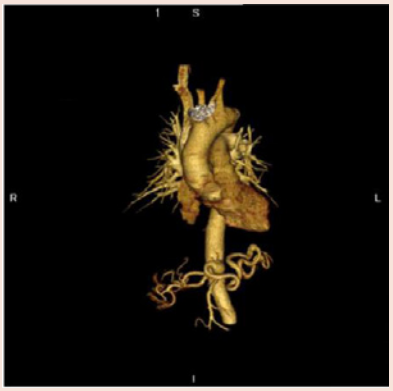

Figure 1a: CT reconstruction of initial celiac artery aneurysm.

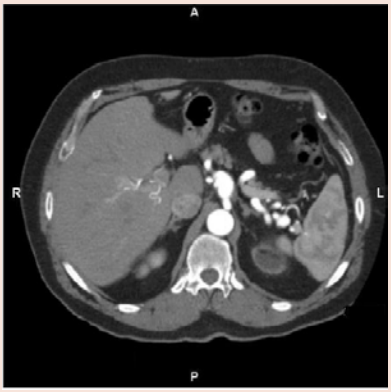

Figure 1b: CT scan (axial view) of initial celiac artery aneurysm.

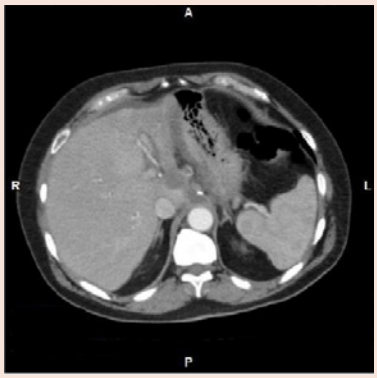

Figure 2a: CT scan (axial view) showing perigastric stranding.

revealed that the distal graft anastomosis was disrupted. The infected graft was removed and the common hepatic artery and the splenic artery were ligated after confirming adequate back bleeding. Fibrotic tissue was encountered around the lesser curvature of the stomach and the first portion of duodenum. Esophago-gastro-duodenoscopy revealed small ulceration along the posterior wall of the first portion of the duodenum, which was communicating with infected fluid collection encountered in the lesser sac. This confirmed the pre-operative suspicion of a possible celiac artery graft enteric fistula. The duodenal ulceration was closed primarily and subsequently covered with an omental patch. Further evaluation revealed an ischemic spleen leading to splenectomy. Intra-operative evaluation of the rest of viscera including the stomach, duodenum, small bowel, large bowel, and liver did not reveal any evidence of ischemia after splenectomy. Intra-operative drains were placed in lesser sac and the splenic fossa.

Post-operatively, the patient's hospital course was complicated by recurrent infectious collection in the lesser sac, which was treated with antibiotics and percutaneous drainage (Figures 3,4). Once he started tolerating regular diet and was found be afebrile, the patient was discharged home on long-term antibiotics and the percutaneously placed lesser sac drain. Post-splenectomy vaccines were administered prior to discharge. The duration of hospitalization was 3 weeks. After 6 weeks of treatment with antibiotics, the percutaneous drain was removed after the patient was noted to have normal vital signs, normal white blood cell count, and no evidence of residual collection in the lesser sac on outpatient computed tomography of abdomen. The patient was lost to follow-up after his 3 month outpatient post-operative visit. At the time of the last follow-up, the patient was

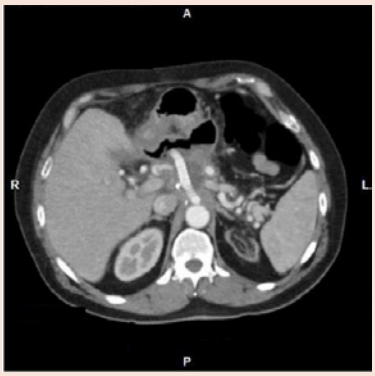

Figure 2b: CT scan (axial view) showing preoperative fluid collection around the prosthethic graft.

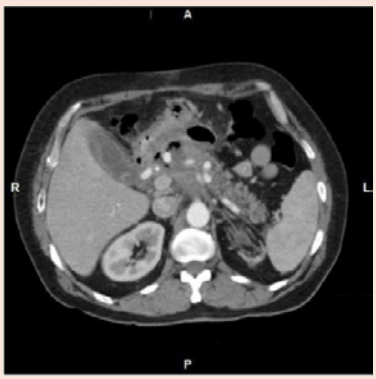

Figure 2c: CT scan (axial view) showing air bubbles within the collection around the graft suggestive of infection.

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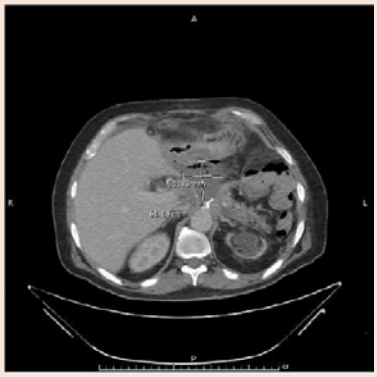

Figure 3: Post-operative CT scan (axial view) showing persistent collection in the setting of fever.

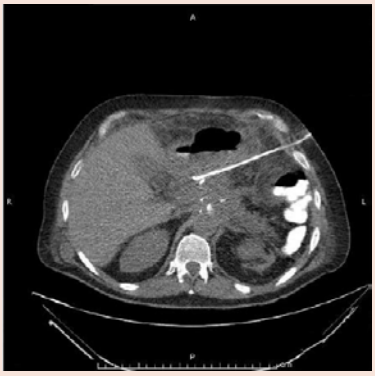

Figure 4: Post-operative CT scan (axial view) showing percutaneous drainage of post-op collection.

tolerating diet well, had normal vital signs, and had normal white blood cell count. An outpatient computed tomography of abdomen performed at 3 month follow-up did not reveal any residual collection in the lesser sac.

Discussion

Since there is a paucity of literature on celiac arterio-enteric fistulas, we will briefly review the well-known entity of aortoenteric fistulas. Concurrently, we will draw parallels between the characteristics of these two clinical entities.

Aortoenteric fistulas are categorized into two categories:

a) *Primary aortoenteric fistulas* occur due to the presence of aortic aneurysm, which could be caused by atherosclerosis or underlying infectious etiology. The incidence of primary aortoenteric fistula is 0.007 per million [6]. The most common location of primary aortoenteric fistula is in the 3rd portion of the duodenum [7].

b) *Secondary aortoenteric fistulas* occur in the setting of previous aortic aneurysm repair. The leading cause of secondary aortoenteric fistulas is prosthetic graft infection [7]. The incidence of secondary aortoenteric fistula is 0.6-2.0 % [6]. The most common location of a secondary aortoenteric fistula is proximal to the prosthetic site [7].

A similar classification may be applied to celiac arterioenteric fistulas. To our knowledge, primary celiac arterioenteric fistulas have not been reported in the literature. This can be explained by the low incidence of celiac artery aneurysms. With the advent of modern diagnostic imaging and increasing use of endovascular treatment of celiac artery aneurysm, this entity may only remain a theoretical possibility. On the other hand, our present case describing secondary celiac arterioenteric fistula after open repair and the case reported by Dinter et al. [8], describing secondary celiac arteriogastric fistula after endovascular repair suggests that secondary celiac arterioenteric fistulas, although extremely rare, can occur after celiac artery aneurysm repair.

The classic clinical presentation of a patient with an aortoenteric fistula is GI bleeding, abdominal pain, and pulsatile abdominal mass [7,9]. These patients usually present with a "herald bleed" in the form of hematemesis and/or hematochezia, which then progresses to hemorrhagic shock [7,9]. In a patient, who presents with an upper or lower GI bleeding, it is essential to determine any known history of aortic aneurysm. The presence of an aortic aneurysm or previous aortic aneurysm repair in this setting should raise the possibility of an aortoenteric fistula.

In our case, the patient presented with symptoms of abdominal pain and fever and subsequently developed a "herald bleed" in the form of hematemesis. We suggest that in a patient presenting with an upper or lower GI bleeding, the physician conducting the interview should include an inquiry about previous splanchnic artery aneurysm repair in addition to an inquiry about previous aortic aneurym repair.

No imaging modality has a high sensitivity and specificity for the diagnosis of aortoenteric fistula. Computed tomographic (CT) evaluation of the abdominal aorta is routinely performed as the initial test if there is a high clinical suspicion for aortoenteric fistula. CT scan findings that are strongly suggestive of a secondary aortoenteric fistula are extravasation of aortic contrast material into the bowel lumen, perigraft gas (>4 weeks after initial repair), focal bowel wall thickening, disruption of aortic wall, and perigraft fluid collection (>3 months after initial repair) [6]. Alternatively, magnetic resonance (MR) imaging of the abdominal aorta may be used but its time-consuming nature is a relative disadvantage in an emergent setting [6,7]. In our case, imaging [CT scan] revealed perigraft gas and perigraft fluid collection and these findings were suggestive of graft infection. The presence of extravasation of enteric contrast into paraprosthetic region is an extremely rare but very specific sign for aortoenteric fistulas [10]. However, this sign was not present in our case.

Esophagogastroduodenoscopy [EGD] should be performed expeditiously to identify the presence of any fistula or extrinsic compression in the 3rd or 4th portion of duodenum [7,11]. However, less than 40% of cases of aortoenteric fistulas are identified via EGD due to characteristic intermittent bleeding pattern [7,11]. In our case, EGD was performed intra-operatively since the diagnosis of celiac arterio-enteric fistula was suspected based on the temporal progression of symptoms from abdominal pain and fever to hematemesis in the setting of perigraft infection. EGD was successful in finding the fistula in the first portion of duodenum in our case.

Once the diagnosis of aortoenteric fistula is suspected, urgent surgical repair is indicated after urgent diagnostic evaluation with CT scan and EGD has been conducted [12]. As soon as the diagnosis of aortoenteric fistula is suspected, broad-spectrum antibiotics providing coverage for gram-positive, gram-negative, and enteric pathogens should be started and continued for at least 6 weeks post-operatively [12]. The main principles of surgical repair of aortoenteric fistula include control of hemorrhage, control of sepsis, and maintenance of distal limb perfusion [13]. This can be accomplished either by the traditional open surgical approach or via an endovascular approach.

There are various options available for open surgical treatment of aortoenteric fistulas like simultaneous graft excision with extra-anatomic bypass, staged extra-anatomic bypass before or after graft excision, graft excision alone, graft excision with in-situ replacement, or primary repair of the graft [14]. The intestinal portion of the fistula can be treated with direct primary repair, segmental resection or proximal diversion [14]. Patients with sepsis, multiple co-morbidities and low life expectancies are possible candidates for endovascular approach since this approach is associated with a lower peri-operative complication rate and lower hospital stay [15]. The infected graft remains in-situ and this increases the risk of infection in the newly placed endovascular graft despite antibiotic therapy [16]. As a result, the short term benefit associated with endovascular graft repair of secondary aortoenteric fistula is negated by the long-term probability of graft infection and sepsis [13]. Another major limitation of the endovascular approach is that it does not include intestinal repair. The management of intestinal portion of fistula requires additional procedures like open repair of intestinal portion or proximal diversion [14].

The mortality for an untreated celiac arterio-enteric fistula is 100%. The case report by Dinter el al. [8], describes a secondary celiac arterio-gastric fistula, which resulted in coil migration into the stomach causing a "herald bleed" and subsequent progression to massive gastrointestinal bleeding leading to death without intervention. Therefore, urgent repair of celiac arterio-enteric fistula is recommended with urgent pre-operative diagnostic evaluation with CT scan and EGD to confirm the diagnosis. The main principles for treatment of celiac arterio-enteric fistula are similar to the treatment of aortoenteric fistula mentioned above. We recommend an open surgical approach for treatment of celiac arterio-enteric fistula.

Control of hemorrhage can be achieved by medial visceral rotation to achieve access to supraceliac aorta for proximal control [12]. Sepsis can be controlled by excision of infected prosthetic graft and thorough debridement of paraprosthetic region [12]. The intestinal portion of the fistula can be repaired primarily [14]. The major difference between open treatment of secondary aortoenteric fistula and secondary celiac arterioenteric fistula is the necessity of bypass in the case of aortoenteric fistula. In the case of celiac arterioenteric fistulas, ligation of the celiac artery stump without revascularization may be attempted once adequate collateral circulation is ensured. In fact, ligation without reconstruction is an acceptable option for mycotic celiac artery aneurysms [2]. In our case, we decided to perform ligation without revascularization due to adequate back-flow from the common hepatic artery and splenic artery initially. The main complications associated with ligation of celiac artery stump are stump rupture, hepatic necrosis, bowel infarction, or splenic infarction [2]. Unfortunately, in our case, splenic ischemia was noted once the intestinal repair was performed. This was possibly due to an error in surgical judgement about the presence of adequate collateral circulation to the spleen after ligation of the celiac artery stump. Subsequently, splenectomy was performed. Revascularization with a prosthetic graft may have been attempted but this raises the issue of placing prosthesis in an infected field. Revascularization with an autologous vein graft may have been another option for restoring splenic perfusion. However, given the degree of splenic ischemia and the length of the case, an intra-operative decision was made to perform splenectomy rather than perform a revascularization procedure. A systemic review of endovascular approach to treat aortoenteric fistula has shown that presence of clinical, laboratory, or imaging signs of infection pre-operatively was predictive of poor outcome (persistent or recurrent infection) after endovascular treatment of aortoenteric fistula in 44% of patients [13]. High risk of persistent infection in this patient deterred us from performing revascularization with another prosthetic graft.

Conclusion

In a patient with a known history of celiac artery aneurysm repair, a high index of suspicion for secondary celiac arterioenteric fistula is required if there is any clinical evidence of GI bleeding due to the high probability of mortality associated with a missed diagnosis. There is a "herald bleed" in the form of hematemesis or hematochezia (depending on the level of fistula) followed by massive GI bleeding manifesting as hemorrhagic shock [8]. Computed tomography is the diagnostic tool of choice for evaluation in patients suspected of celiac arterioenteric fistula. CT features suspicious for celiac arterioenteric fistula are similar to aortoenteric fistula and include presence of ectopic gas, extravasation of aortic contrast into paraprosthetic space, and extravasation of enteric contrast into paraprosthetic space [6]. EGD must be performed to confirm the diagnosis. Treatment for secondary celiac arterioenteric fistula should include graft excision followed by celiac artery stump closure followed by primary repair of intestinal portion of the fistula. Revascularization may not be necessary if there is adequate collateral circulation.

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