

PSEUDO-ANEURYSM OF DISTAL AORTIC STUMP – A MANIFESTATION OF AORTOENTERIC FISTULA

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ABSTRACT

Background: Secondary aortoenteric fistula (AEF) is rare but devastating complication of abdominal aortic reconstruction surgery for aortic aneurysm. Most patients presented with acute upper gastrointestinal bleeding. CTA abdomen is usually performed to attain the diagnosis. However, AEF has invariable appearances on CT where mostly are non-specific and similar to perigraft infection (PGI). A case of secondary AEF following multiple series of aortic surgery, with CT manifestation of recurrent pseudo-aneurysm of aortic stump, is presented.

Case Presentation: 65 years old man who had undergone series of aortic reconstructive surgery for infra-renal abdominal aortic aneurysm presented acutely for hematemesis and per rectal bleeding. Initial CTA abdomen revealed pseudoaneurysm of the distal aortic stump, which was successfully embolized with coils. However, the symptoms recurred and repeated CTA showed new pseudo-aneurysm but no typical findings of AEF. Emergency laparotomy confirmed AEF at duodenojejunal junction that was repaired with primary closure. Unfortunately, patient died 3 days later due to multiple post-operative complications.

Conclusion: Presence of pseudo-aneurysm with background of perigraft infection should raise the suspicion of AEF, especially in patients with history of aortic intervention that presented with gastro-intestinal bleeding. CTA abdomen is invaluable in providing the diagnosis of AEF despite its variable manifestations.

MeSH Keywords: Aorto-enteric fistula, pseudo-aneurysm, aortic stump, perigraft infection,

CTA abdomen, endovascular aneurysmal repair, coil embolization, aortic explantation, acute upper gastrointestinal bleeding.

Introduction:

Secondary aortoenteric fistula (AEF) is rare but devastating complication of abdominal aortic reconstruction surgery for aortic aneurysm. Most patients presented with acute upper gastrointestinal bleeding (UGIB). CTA abdomen is usually performed to attain the diagnosis. However, AEF has invariable appearances on CT where mostly are non-specific and similar to perigraft infection (PGI). A case of secondary AEF following multiple series of aortic surgery, with CT manifestation of recurrent pseudo-aneurysm of aortic stump, is presented.

Case Report:

65 years old man with background history of diabetes mellitus and hypertension was rushed to our emergency department for acute hematemesis and per rectal bleeding for the past 4 days. He had past surgical history of EVAR for an infra-renal abdominal aortic aneurysm 2 years ago, which was complicated with prolonged history of infected aortic endograft, leading to aortic endograft explantation and left axillo-bifemoral bypass surgery. Clinically, he was pale and digital rectal examination revealed hematochezia. An urgent oesophagodoudenoscopy (OGDS) showed no significant findings.

CT angiography of abdominal aorta was then performed without oral or rectal contrast instillation. Plain phase of CTA showed presence of intraluminal hyperdensity within sigmoid colon. However, no increased in attenuation value and size of the intraluminal hyperdensity in the subsequent

arterial, portovenous, and delayed phases. Strut noted at the distal aortic stump with extensive peri-aortic enhancement and thickening in keeping with PGI.

Sigmoidoscopy done the next day confirmed the presence of mixed fresh and old blood within the sigmoid colon but no demonstrable active bleed was seen. An immediate repeat CTA abdomen showed active contrast extravasation from the distal aortic stump into the retroperitoneal space, adjacent to D4 in keeping with pseudo-aneurysm.

Initial aortogram performed using 5Fr pigtail catheter showed no demonstrable abnormality. Selective angiogram of the distal stump was then performed using Burn 5Fr catheter and Boston microcatheter, which confirmed the presence of pseudoaneurysm at anteroinferior aspect of the distal aortic stump. Coil embolization was performed using 4 Boston interlock coils (two 14mm x 50cm, one 4mm x 15cm, and a 5mm x 15cm). Post coiling angiogram run showed no contrast opacification of the pseudo-aneurysm sac in keeping with complete embolization.

Patient was well up until 3 days later where he had sudden onset of UGIB with associated hypotensive episode. Urgent CTA abdomen showed new pseudo-aneurysm within the distal to the aortic stump, anterosuperior to the previously embolized pseudo-aneurysm. No definite evidence to suggest AEF.

Emergency exploratory laparotomy confirmed presence of aorto-enteric fistula at duodenojejunal junction, measuring 0.5 x

0.5cm, which was repaired with primary closure with 3 0 Vicryl suture. The distal aortic stump was friable with strut noted within. The strut was removed while the stump was debrided and over sewn using 3 0 Prolene suture. Intra-operatively, his sigmoid colon was noted to be dusky. However, the patient was sent back to intensive care unit for stabilization due to prolonged surgery and was planned for second look laparotomy after 24 hours. Unfortunately, patient's condition deteriorated due to severe sepsis and multi-organ failure. He ultimately succumbed to death 3 days later.

Discussion:

Aorto-enteric fistula (AEF) is defined as abnormal communication between the aortic lumen and the gastro-intestinal tract. There are two major etiologies that have been postulated in literature for the development of AEF. First etiology is mechanical injury to the bowel, which leads to focal necrosis causing bowel wall erosion. Bowel injury can be caused by multiple factors namely from dissection during surgery, direct pressure of anastomotic pseudo-aneurysm, and direct pulsatile pressure by the adjacent graft. Second etiology is infection as a result from contamination and post-operative bacteremia of any cause [1]. Regardless of these etiologies, adherence of aorta or aortic graft to the bowel segment is vital for a fistulous tract to form in between [2,3,5,6].

AEF is classified as primary and secondary. A primary AEF is caused by spontaneous erosion whereby secondary AEF happens as a complication of endovascular interventions or open surgery [1]. Secondary AEF has a median age of 65 years old and men

are more commonly affected compared to women [1].

The reported incidence of secondary AEF is higher in emergency settings (14%) as compared to elective case (1%) of aneurysm repair [1,4]. Elective abdominal aortic aneurysm (AAA) resection is the most known surgery to result in secondary AEF. Other procedures that may give rise to this complication include aortic replacement or bypass surgery in case of aorto-iliac occlusive disease, ruptured AAA resection, and graft stenting of AAA [1,4]. The interval duration for AEF to develop in a patient range from as fast as 2 days up until 26 years. The reported median interval is between 24 to 47 months [1].

The patient's latest surgery prior to his presentation was exploratory laparotomy for PGI, which is approximately 1 month in duration. The short interval duration of AEF formation in this patient is likely due to his extensive history of aortic procedures ranging from EVAR, graft stenting, and ultimately aortic graft explantation and bypass surgery. Furthermore, the presence of strut within the distal aortic stump is likely the cause of the recurrent PGI in which it provides potential conduit for bacterial colonization; consequently, leading to AEF.

As in term of the location, AEF found in this patient at duodenojejunal junction corresponds to the most predisposed location for AEF formation; which are D3 and D4 [2,4]. These are likely due to their retroperitoneal location and its close proximity with the graft. Other possible fistulous locations include the stomach and appendix [1,4].

The most devastating manifestations

of AEF are hematemesis, malena, and even shock. However, not every patient with AEF presents with evident gastrointestinal bleeding. Some of them may present with abdominal pain, sepsis, fever, or back pain [1].

OGDS has a low diagnostic accuracy of 30% in evaluating AEF [1]. However, the main role of OGDS is basically to exclude other possible causes of the gastrointestinal bleed [5-7].

CTA abdomen is regarded as the best imaging modality for stable patients with suspected AEF due to its easy-availability and its high sensitivity (94%) and specificity (85%) in diagnosing perigraft infection with or without AEF [8,9]. However, AEF has various appearances in CT. The most definite but rare features of AEF are not demonstrated in this patient's CTA abdomen. These definitive features inclusive of intraluminal contrast extravasation into bowel, extraluminal gastrointestinal contrast leakage into the peri-aortic space, and presence of aortic graft in the bowel lumen [10,11]. However, CTA of this patient are able to show non-specific features of AEF, similar to PGI; which are pseudoaneurysm formation and extensive distal stump para-aortic soft tissue thickening with no demonstrable fat plane with the adjacent duodenum. Other non-specific features of AEF/PGI are presence of gas collections within or surrounding the graft, focal bowel wall thickening adjacent to aortic graft, and focal disrupted aortic wrap calcification [8-12].

AEF can either be managed by conventional surgery or endovascular repair. Conventional surgery is proven to have high mortality and morbidity rate in previous literatures. The surgery consists of graft

explantation followed by primary or secondary axillo-bifemoral bypass and in situ reconstruction of the aorta. Endovascular intervention is a new method of rapid bleeding control and restoration of peripheral perfusion in hemodynamically unstable patients. This technique is considered as temporizing procedure prior laparotomy and part of long-term palliative care in high-risk surgical patients [13-15]. As for example, this patient did benefit from initial successful coil embolization as evident by immediate transient clinical improvement post-procedure. However, the development of new pseudo-aneurysm in short duration indicates the fragility of the distal aortic stump and infectiousness of the perigraft / peri-aortic tissues.

Conclusion:

In conclusion, AEF is a lethal complication in post EVAR patients with mortality of almost 100%. Presence of pseudo-aneurysm with background of PGI should raise the suspicion of AEF. This is especially true in patients with history of aortic intervention that presented with gastrointestinal bleeding. OGDS almost always yielded negative result. CTA abdomen is invaluable in providing the diagnosis. Hence, familiarization with varieties of CT appearances in AEF is warranted for prompt and accurate diagnosis.

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Figure Legends:



Figure 1: Selected axial image of CTA Abdomen (arterial phase)



Figure 2: Selected sagittal image of CTA Abdomen (arterial phase) with MIP.

Figure 1 & 2: CTA Abdomen shows presence of aortic stump leak (adjacent to D4 of the duodenum) in keeping with pseudo-aneurysm. Note that the strut within the distal aortic stump with extensive surrounding soft tissue thickening.

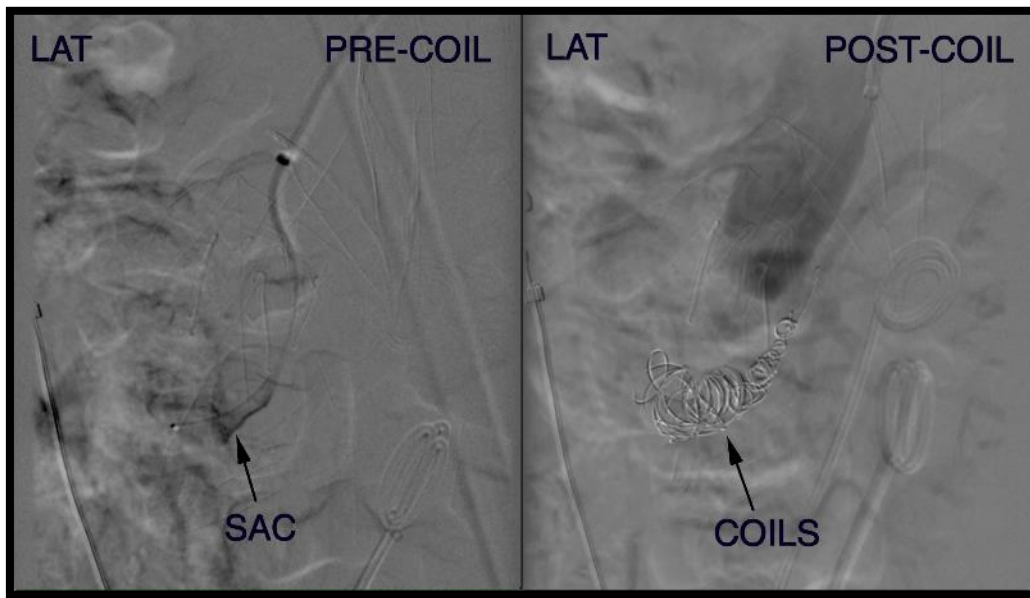


Figure 3: Pre-coiling angiogram demonstrates pseudo-aneurysm sac distal to the strut of the distal aortic stump. Post-coiling angiogram shows multiple coils within the pseudo-aneurysm sac.



Figure 4: Selected axial image of CTA Abdomen (arterial phase).

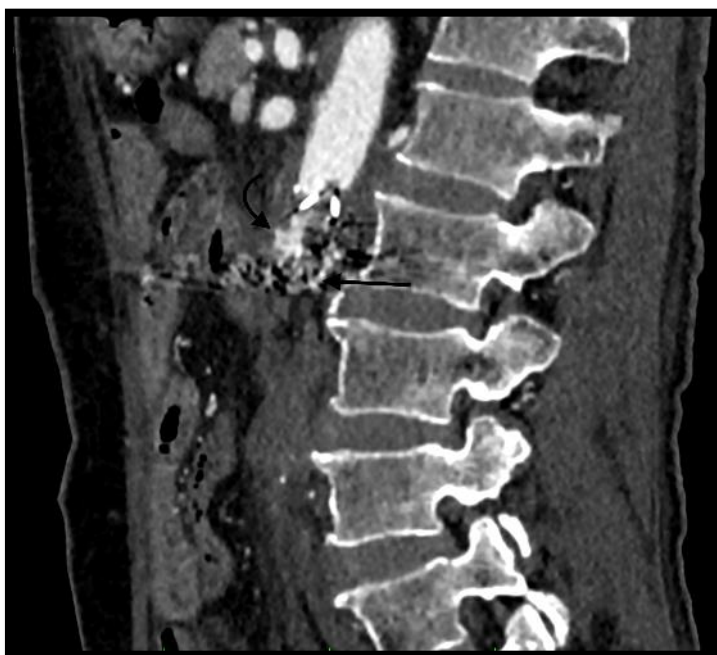


Figure 5: Selected sagittal image of CTA Abdomen (arterial phase).

Figure 4 & 5: Repeated CTA Abdomen 3 days later shows presence of new pseudo-aneurysm (elbowed arrow), distal to the aortic stump, and anteriosuperior to the previously embolized pseudo-aneurysm (straight arrow).