Endovascular Treatment of Ruptured Secondary Aortoenteric Fistula 10 years after Open Repair of AAA. Case Report and Literature Review.

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Abstract

Secondary Aorto Enteric Fistula, most commonly occur after previous aortic surgery, either open or endovascular and constitute surgical emergency, requiring prompt management.Hence,is reported a case of a 63 years old male with secondary aortoenteric fistula rupture into bowel treated endovascular while mid-term results, along with a thorough literature review

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Key Clinical Message

Secondary Aorto Enteric Fistula, most commonly occur after previous aortic surgery, either open or endovascular and constitute surgical emergency, requiring prompt management. Hence,is reported a case of a 63 years old male with secondary aortoenteric fistula rupture into bowel treated endovascular while mid-term results, along with a thorough literature review.

Keywords: Aortoenteric fistula, aortoenteric rupture, aortic pseudoaneurysm, aortic infection and endovascular aneurysm repair.

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INTRODUCTION

Secondary aortoenteric fistula (SAEF) is a rare but well-known complication after aortic aneurysm repair, which was first described in 1998 by Norgren.[1] The diagnosis remains a challenge, as the suggested diagnostic procedure is the abdominal contrast CTA, which provides vague and non-specific findings. This article presents a case report and a literature review of a SAEF rupture and concludes the main points of diagnosis and treatment of this uncommon clinical entity.

CASE PRESENTATION

A 63-year-old male was presented to the Emergency Department in haemo-dynamic shock (BP 85/55mmHg, HR 117bpp, anuria).

Patient symptoms began six days ago, with abdominal pain, localized in the lumbar area. The pain appeared suddenly and was followed by an episode of haematochesia. Ever since, the patient reports repeated bleedings per rectum (PR) and a fever up to 38.9 ^{o}C degrees.

Prior medical history included hypertensive cardiopathy, diverticulitis, open repair of abdominal aortic aneurysm (AAA) 10 years ago and plastic reconstruction of postoperative abdominal midline hernia a year ago.

After primary fluid resuscitation an abdominal CT scan was ordered, in which a mass was depicted alongside distal aortic anastomosis of the previous tube synthetic graft. (Figures 1- 2).

In the contrast enhanced CT scan, active extravasation is visible at the same level of the aortic graft into a sac containing attenuating clot and ectopic gas. (Figure 3).

The patient was, thereby, lead to the OR, where a ruptured distal anastomosis of his previous AAA repair was found indeed and under direct Fluoroscopy an endovascular aortic aneurysm repair (EVAR) with the use of a bifurcated stent graft placement over the rupture occurred.

Intraoperative completion angiography confirmed satisfactory placement of the stent and no leaks or endoleaks were identified along with simultaneous hemodynamic stability of the patient.

Five days postoperative, a repeated CTA was done, exhibiting satisfactory placing of the stent, total sealing of the rupture with no signs of leakage at the periaortic region or inside the bowel. (Figure 4)

The patient's postoperative period was uneventful and in seventh day postop he was driven again to the OR where a wide surgical debridement with sigmoidectomy and Hartmann procedure took place.

Postoperative period was uneventful, infection markers normalized, and the patient presented no signs of fatigue or fever.

The patient recovered well and was discharged 16 days postoperatively, after the consultation of the Infections Disease Experts Committee, for a six weeks protocol of antibiotic treatment.

Three months later returned to our clinic for a successful restoration of the bowel continuity and at 6 and 12 months follow up the patient remains in extraordinary condition with no reported complications and completely regression of pseudoaneursym sac (Figures 5, 6).

Therefore he remains well with no signs of fatigue or fever, normal infection markers and has totally returned to his previous activities while he is monitored outpatient regularly.

DISCUSSION

Aortoenteric fistulas (AEF) constitute a rare clinical entity, concerning a pathological connection between the aorta and the gastrointestinal tract. Although their incidence barely reaches 4% (between 1.6 and 4%), they are associated with high mortality rates (24% to 45.8%).[2, 3]

They are divided into two major categories, the Secondary AEF (SAEF), which most commonly occur after previous aortic surgery, either open of endovascular, and the Primary AEF (PAEF), that take place spontaneously without any previous intervention in the aorta and are even rarer (incidence <0.07%)[4].

High-risk patients of developing SAEFs include those who undergo emergent surgery for a ruptured aneurysm, have post-operative complications such as reoperation or bowel injury, and those with endoleaks or stent migration.[5]

Most commonly, an AEF presents with an initial or multiple "herald bleedings", meaning episodes of transient, seemingly self-limiting and minimal lower GI bleeding. A catastrophic bleed follows, resulting in significant hemodynamic instability, requiring urgent surgical repair. Other clinical signs and symptoms might be sepsis (44%), abdominal pain (30%), back pain (15%), groin mass (12%), and abdominal pulsatile mass (6%).[6]

Clinical symptoms are very important in diagnosis of an aortoenteric fistula, because of the lack of high sensitivity and specificity of a single imaging modality. In previous reports authors suggested the diagnostic triad of pain, sepsis, and gastrointestinal bleeding, although the septic element is not always present in a SAEF.[8]

In our case, the patient presented with septic shock. This was explained by the infected pseudoaneurysm, which was the cause of the fistula. The previous medical history should arise the suspicion of such a diagnosis, creating the question of the best diagnostic imaging test.

Upper endoscopy is useful to exclude other sources of bleeding, such as peptic ulcer disease, but a normal endoscopy result never rules out AEF. When in suspicion of a SAEF, CT scan is considered the work-horse exam, although literature reports variable sensitivity and specificity.[8] The most important imaging finding in an AEF is ectopic gas in the aortic lumen or in direct contiguity to the aortic lumen. In the most obvious cases, a direct tract of gas can be traced directly from the involved bowel loop towards the aorta.[7] However, while highly suggestive; the presence of gas is not completely specific. For example, perigraft soft-tissue edema, fluid, and ectopic gas may be normal CT findings immediately after surgery.[9] However, after 2-3 months, identification of any ectopic gas should be considered a sign of perigraft infection with the possible presence of AEF until proven otherwise.

Other CT findings that are suggestive of both perigraft infection and aortoenteric fistula include pseudoaneurysm, effacement of the periaortic fat, tethering of a bowel loop immediately adjacent to the aorta, disruption of the aortic wall or a graft or significant graft migration.[7]

The most specific sign in AEF, direct extravasation of contrast from the aortic lumen into a intestinal loop, is especially rare to identify on CT. Similarly, the leakage of enteric contrast directly into the periaortic space is a highly specific sign, but extremely rare. Notably, in a series by Hagspiel the extravasation of contrast from the aorta into the bowel lumen was present in only 11% of cases.[14]

Digital Subtraction Angiography (DSA) is frequently requested for further evaluation of gastrointestinal bleeding. However, in most patients with bleeding in the gastrointestinal tract, DSA offers few, pointing of to be a poor diagnostic tool.[14] On the other hand CTA is proven to be a helpful tool for identifying disruption of the aortic wall, outline the a pseudoaneurysm, or demonstrating the presence of an AEF by describing direct extravasation from the aorta into the gastrointestinal lumen.[10-11]

The nonspecificity of these features is responsible for a considerable overlap with a variety of other disorders, with perigraft infection being the most important, as it can appear identical to a fistula. Other entities which intrigue the diagnostic procedure include aortitis, mycotic aneurysms, and perianeurysmal fibrosis, all of which can demonstrate periaortic inflammation, fluid, or soft tissue.[7, 12, 13]

Intramural gas and aortic wall thickening in a patient with positive blood cultures should raise suspicion of the presence of infectious aortitis. Similarly, when a patient is presented with a saccular aneurysm of the aorta accompanied with clinical evidence of sepsis, the surgical team should act prompt to treat the infected aortic aneurysm before it ruptures.[7]

As noted in the beginning, a secondary aortoenteric fistula occurs most commonly after an aortic aneurysm reconstruction. Whether this complication occurs more frequently after an open or an endovascular repair

remains open to interpretation.[16, 17]

Considering that arterial suture line appears to be the main predisposing factor in open repair, endovascular treatment of AAA was initially thought to confer little to no risk of AEF. However, persistent endoleak, with the continued presence of blood flow into the aneurysm sac and aneurysm sac enlargement, represents a significant complication of endovascular aneurysm repair.[18]

Further, degeneration of the aneurysm neck causes graft migration with proximal endoleak and presence of pulsatile blood flow into aneurysm sac which eventually will lead to continued aneurysm expansion and rupture or subsequent bowel erosion and creation of AEF.[19]

Identification of impending or contained rupture is critical because these patients are at risk for frank rupture but can generally benefit from a more thorough preoperative assessment, followed by urgent surgery.[20]

Regardless the initial reconstructive surgery, when a SAEF is present, traditionally is managed by graft explantation, wide debridement of the infected tissues, infrarenal aortic stump ligation, and extra-anatomic revascularization with axillo-bifemoral bypass.[21]

This former gold-standard procedure has been doubted, as it hides the risk of aortic stump blow-out syndrome, specifically in long term follow up. As a result, in situ bypass grafting using homografts, allografts, prosthetic or vein grafts was developed. [23-25]

In a series published by Kakkos et al, open surgery had higher in-hospital mortality (246/725, 33.9%) than endovascular methods (7/89, 7.1%, $p<0.001$). The reduced postoperative mortality after endovascular surgery indicated that expedient haemostasis achieved with former methods without any further insults is probably all that is needed in this patient population with haemmorhagic shock. The statistical difference, however, mostly disappeared during the first 18-24 months after the procedure.[22]

Most common causes of death in short-term post-operative period were irreversible shock, cardiac arrest, bleeding – either from stump blowout or homograft rupture- sepsis, MODS and GI complications including leak from the GI repair. During long-term follow up bleeding (recurrent AEF, stump blowout, disruption of the proximal anastomosis or homograft rupture), sepsis MOF or coronary syndromes were noted as the commonest causes of death.

CONCLUSION

Secondary aortoenteric fistulas can be challenging to diagnose as the CT features of aortoenteric fistulas can be quite subtle. In the same time their handling and treatment remain a point of controversy, with the scale leaning towards the endovascular techniques. The above facts in combination with the high mortality of this clinical entity highlight the significance of doctor's awareness, as an early diagnosis can spare the patient from a lethal outcome.

Conflict of Interest

None

Author Contribution

IS: Senior surgeon managed the patient and senior manuscript author. EK: Junior doctor and primary manuscript author. HEZ: Senior surgeon managed the patient. FT: Junior doctor managed the patient. LP: Junior doctor managed the patient. AK: Junior doctor managed the patient. EM: Senior author involved in editing the manuscript. GL: Senior surgeon managed the patient. DL: Senior author involved in editing the manuscript. KT: Senior author managed the patient. AR: Overall senior surgeon managed the patient and involved in editing the manuscript.

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