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CASE REPORT

Aortoduodenal Fistula from Duodenal Stenting for Malignant Gastric Obstruction

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Introduction: Aortoduodenal fistula (ADF) is a rare cause of upper gastrointestinal (GI) bleeding and is usually fatal without intervention. A high index of suspicion is required to identify and successfully manage this condition.

Report: Three cases of ADF following duodenal stent insertion for gastric outflow obstruction secondary to metastatic adenocarcinoma are presented. All presented with upper GI bleeding and underwent emergency percutaneous endovascular aortic stent graft repair (EVAR), with temporary aortic balloon occlusion in one case. All were successful in achieving haemostasis. The first case, although initially complicated by acute stent thrombosis of the right iliac limb, was discharged to a rehabilitation facility and survived for two years. The second patient died two days after the procedure from sepsis related complications. The third was discharged home six days post-procedure with a three month follow up.

Conclusion: A high suspicion for ADF in patients with previous duodenal stents is required for prompt diagnosis and management. EVAR may increase short to midterm life expectancy.

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INTRODUCTION

Aortoduodenal fistula (ADF) is an uncommon but catastrophic cause of gastrointestinal (GI) bleeding. Secondary ADF is a recognised rare complication of endovascular abdominal aortic repair (EVAR) with an incidence of 0.46%—3.9%.[1] Duodenal stent insertion is considered efficacious for the palliative management of obstructive malignancy, and bleeding secondary to erosion of enteric stents has been documented.[2] However, secondary ADF from duodenal stents is rare, with one case report published.[3] Three cases treated by endovascular means are presented; although the treatment was technically successful, morbidity and mortality was high.

CASE REPORT

Case 1

A 51 year old man presented with haematemesis and collapse, seven months after the insertion of a duodenal WallFlex 120 × 22 mm stent (Boston Scientific, Marlborough, MA, USA) for gastric outlet obstruction for metastatic colorectal adenocarcinoma diagnosed five years earlier and previous chemoradiotherapy. On presentation, the patient was in shock (blood pressure 95/60 mmHg), with a distended abdomen and haemoglobin of 7.2 g/L, despite having received three units of packed red blood cells (pRBC). Intravenous (IV) fluid resuscitation with prophylactic antibiotics was administered. Triphasic computed tomography angiogram (CTA; Fig. 1) identified locules of gas within the retroperitoneal para-aortic soft tissue, in keeping with a fistulous communication with the duodenum; CTA did not identify active bleeding. The patient was transferred to the interventional endovascular suite for emergency EVAR.

Bilateral percutaneous femoral access was obtained. Angiography identified aortic narrowing with thrombus at the level of the duodenal stent without pseudoaneurysm. An occlusion balloon was placed within the thoracic aorta for 10 minutes to reduce hypotension. A 23 × 12 × 12 GORE bifurcate stent graft (WL Gore, Flagstaff, AZ, USA) with an iliac artery limb extension (16 × 12 × 10) on the left was implanted (Fig. 2).

Day 1 post-operatively the right iliac limb occluded resulting in acute limb ischaemia, probably due to limb compression within a narrow aorta. Graft thrombectomy, four compartment fasciotomy, and bilateral balloon

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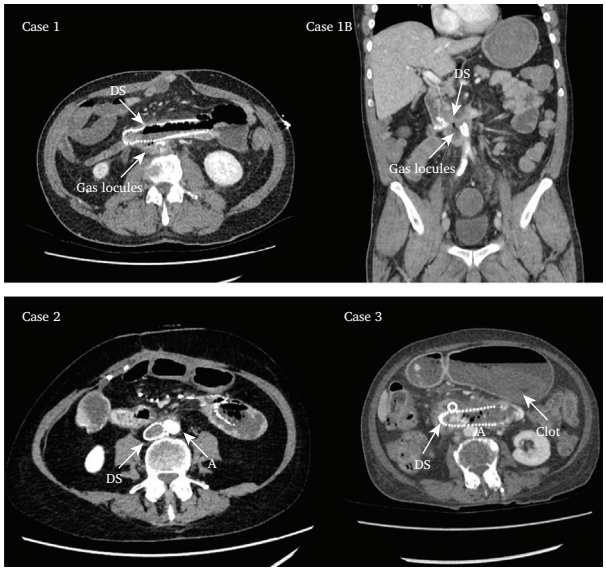

Case 1; Gas locules are visualised within the para-aortic soft tissue immediately adjacent to the duodenal stent (DS).
Case 1 B; The second image shows gas locules within retroperitoneal para-aortic soft tissue.
Case 2; A duodenal stent is seen abutting the infrarenal aorta with loss of the plane between the aorta and duodenum.
Case 3; A duodenal stent is seen abutting the aorta and second arrow indicating clot in the stomach.

Figure 1. Computed tomography angiography images of the three cases.

expandable stents were placed in the contralateral gate and right iliac limb. Post-operative issues included pain, physical deconditioning, and concerns of stent graft infection due to ongoing elevated inflammatory markers, treated empirically with piperacillin—tazobactam. The patient was discharged after five weeks to rehabilitation with lifelong oral moxifloxacin and died at an external facility. His cause of death is not known to the authors; his overall survival was two years.

Case 2

A 59 year old woman presented two years after duodenal WallFlex 120 × 22 mm stenting (Boston Scientific) for symptomatic gastric outlet obstruction due to metastatic small bowel adenocarcinoma. She presented following collapse and frank bleeding into her ileostomy bag. Previous treatment included proctocolectomy with ileostomy for ulcerative colitis in the 1970s, followed by small bowel resection and adjuvant chemoradiotherapy in 2004.

On presentation, the patient was normotensive with a haemoglobin of 11.5 g/L and a temperature of 38.3oC. IV antibiotics were administered and an initial triphasic CTA showed no contrast extravasation into the duodenum (Fig. 1). That evening, she became haemodynamically unstable with further fresh GI bleeding. Urgent transfer to a tertiary facility was arranged for oesophagogastroduodenoscopy (OGD). Despite resuscitation, the patient had a pulseless electrical activity arrest requiring 30 seconds of cardiopulmonary resuscitation. Return of spontaneous circulation was achieved, and vasopressors and pRBCs were given. At OGD a large blood clot was seen in the distal duodenum, but no active bleeder was visualised, raising concern for a fistula. CTA showed a duodenal stent abutting the infrarenal aorta with loss of the aorto-enteric

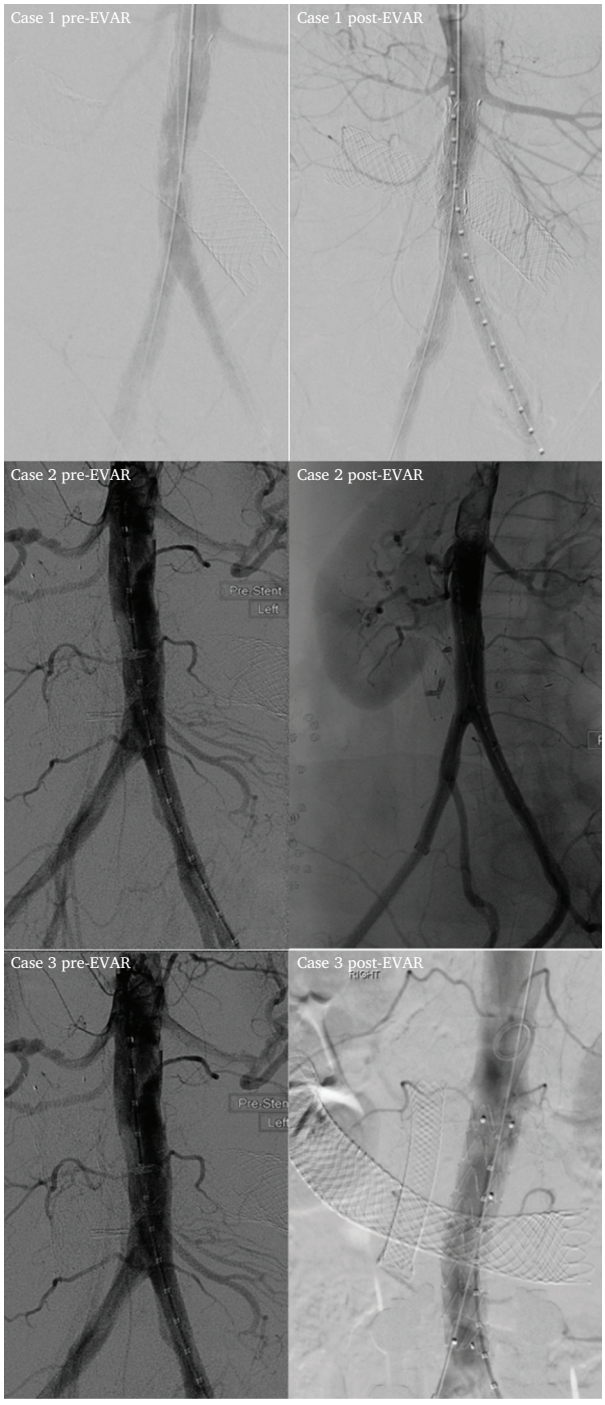

Figure 2. Intraprocedural images of the three cases before and after endovascular aortic stent graft repair.

plane when compared to recent imaging. The patient proceeded to angiogram for EVAR.

Bilateral percutaneous femoral access was used and no pseudoaneurysm was seen. A 23 × 33 mm GORE aortic cuff was inserted to cover the aorta at the stented level (Fig. 2).

The patient's temperature was 38.5oC on day one, with vasopressor support required. Blood cultures from presentation grew *Staphylococcus aureus*, *Enterococcus faecium*, and *Candida albicans*. Despite targeted antimicrobials and

vasopressors, the patient died on day three from cardiac arrest secondary to sepsis and disseminated intravascular coagulation.

Case 3

A 73 year old woman presented following duodenal stent insertion two months previously for gastric outlet obstruction on a background of pancreatic adenocarcinoma diagnosed one year before, previous chemoradiotherapy, and common bile duct stent. She presented with large volume haematemesis and circulatory collapse. Two units of PRBCs and active resuscitation improved the initial haemoglobin of 6.4 g/L to 8.3 g/L. Triphasic CTA identified a duodenal stent eroding the aorta, with active blush and large gastric thrombus (Fig. 1). The patient proceeded to the interventional suite for EVAR.

Bilateral percutaneous femoral access was obtained and no pseudoaneurysm was seen. A 20 × 20 × 80 mm iliac limb (Medtronic, Minneapolis, MN, USA) was deployed (Fig. 2).

Post-operatively, vasopressors and IV piperacillin—tazobactam antibiotics were given. Admission blood cultures grew *Klebsiella pneumoniae* and *Escherichia coli*. Discharge was on day eight with lifelong oral moxifloxacin and she was well at three months.

DISCUSSION

ADF is a rare cause of upper GI bleeding and is fatal without intervention. A high index of suspicion is required to identify and successfully manage this condition. The use of duodenal stents for palliative symptomatic relief of malignant outflow obstruction is well documented, and late complications of bleeding, perforation, and stent migration are recognised.[2] ADF secondary to duodenal stenting is an exceedingly rare complication, with only one case report describing a GI bleeding presentation on a background of malignancy with duodenal stenting eight years previously.[3] Angiography demonstrated an aortic pseudoaneurysm extending to the stent, without extravasation. Aortic endografting was successful. This previously reported patient was then stable at six months on lifelong oral antibiotics.

Diagnosis relies on CTA or OGD.[4] CT may show gas within or next to the aortic lumen, perigraft air, or loss of fat plane between stent and duodenum.[4] Clear fistulous communication or presence of contrast extravasation is not required for diagnosis, and delay may increase mortality. OGD may identify erosions through enteric mucosa, or thrombus on the duodenal wall allowing prompt identification of the fistula site. If thrombus is identified, an urgent surgical opinion should be sought.[4]

Historically, open abdominal aortic repair was recommended for the surgical management of ADF from non-malignant causes as aggressive debridement was necessary for cure.[5] Mortality and morbidity rates from EVAR are lower; in hospital mortality 7.1 *vs.* 33.9%.[5] However, lack of debridement means cure cannot be achieved and antimicrobial therapy is required to suppress infection.[6] There is no consensus on optimal duration of therapy for graft infection.[6]

A systematic review[7] established an association between EVAR for aorto-enteric fistula and infection. They reported a 44% incidence of persistent or new infection at 13 month follow up,[7] with signs of sepsis pre-operatively being associated with poorer outcome. As definitive open surgical repair confers long term benefit from reduced rates of infection and re-bleeding, EVAR is a temporising measure to prolong lifespan.[6] In the cases described herein, open surgery is contraindicated. Palliation prior to EVAR was considered in these cases and the decision to proceed was made in conjunction with a shared care model to extend life.

A general anaesthetic for emergency EVAR is recommended for airway management in the context of haematemesis. An aortic occlusion balloon may be used for intra-operative circulatory collapse, as in case 1.

The choice of graft for the EVAR can be challenging given the small diameters of the vessels concerned and difficulty visualising the point of bleeding. In case 1, a bifurcated graft was placed because the infrarenal aorta was too short for the shortest tube graft and there was no confidence that it would cover the defect with a cuff; unfortunately, this resulted in thrombosis. A tube graft may be sufficient; bifurcated grafts take longer to insert, and the risk of limb thrombosis is higher in the presence of infection, small distal aortic diameter, and prothrombotic state associated with malignancy. The authors suggest that balloon expandable stents in the proximal iliac limbs may reduce this risk if the aorta is small calibre.

Conclusion

ADF is a rare but life threatening cause of GI haemorrhage that requires immediate diagnosis and management and should be suspected in patients with duodenal stents inserted for malignancy. This case report highlights the utility of EVAR for ADF to achieve haemostasis in a patient group with limited prognosis, infection, and comorbidities. Although not without risk, endografting may prolong life expectancy.

CONFLICTS OF INTEREST

None.

FUNDING

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