CASE REPORT

Emergent interventional approach for aortogastric tube fistula with massive gastrointestinal bleeding

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SUMMARY

This report describes the successful endovascular treatment of a rare case of aortogastric tube fistula with massive gastrointestinal haemorrhage. The patient's history included oesophageal reconstruction for oesophageal carcinoma using a gastric tube. Emergent angiography revealed extravasation from the thoracic aorta into the thinner aortogastric tube fistula. A microcatheter was inserted into the aortogastric tube with the aortic approach for embolisation with a mixture of *n*-butyl cyanoacrylate and iodised oil to enable fluoroscopic visualisation. Aortography confirmed the complete absence of extravasation after embolisation, after which a stent graft was placed. This procedure demonstrated that transcatheter embolisation of the aortogastric tube fistula was possible, and that the technique can be used as an emergency option. CT imaging was performed 21 days after the procedure, and revealed no trace of extravasation or inflammation. There were no complications during the 14 months following the endovascular treatment; the patient remains in stable condition.

BACKGROUND

Aortogastric fistula is a very rare condition, particularly in association with gastric tube complications following oesophagectomy.^{1–2} Such injuries to the aorta are rarely observed, and commonly result in rapid exsanguination and a high mortality rate.^{1–3} Twenty-one cases of aortogastric fistula have been reported in the literature, and only one patient survived after undergoing surgery and endovascular stent graft placement.^{1–2} Immediate intervention is the sole option for saving patients with this condition.² We report the successful repair of postoesophagectomy aortogastric tube fistula via embolisation with *n*-butyl cyanoacrylate (NBCA) mixed with iodised oil (hereafter, NBCA/iodised oil) and stent graft placement.

CASE PRESENTATION

A 72-year-old woman underwent transhiatal subtotal oesophagectomy 14 years ago for squamous cell carcinoma of the distal oesophagus. Isoperistaltic gastric tube interposition with cervical oesophagogastric anastomosis was performed in the posterior mediastinum. The patient discontinued follow-up 9 years postoperatively, and experienced a sudden onset of massive haematemesis, gastrointestinal bleeding and critical haemorrhagic shock (stage 3 hypovolemic shock) 14 years postoperatively. The patient was immediately admitted to our emergency department.

INVESTIGATIONS

Critical haemorrhagic shock was treated with blood transfusion and infusion solution. Endoscopy revealed the cause of bleeding to be a peptic ulcer, but haemostasis could not be achieved. Emergency contrast-enhanced CT in the arterial phase showed a well-enhanced lesion in the reconstructed gastric tube. Gastrointestinal bleeding was suspected to have resulted from the aortogastric tube fistula (figure 1).

TREATMENT

Transcatheter arterial embolisation (TAE) was selected, given the unstable circulatory dynamics of the patient and unavailability of stent grafts at the time. Diagnostic angiography and TAE were performed under local anaesthesia. After access was obtained via the femoral artery, a 4 Fr catheter (Shepherd Hook, Medikit, Japan) was placed into the descending thoracic aorta (figure 2A). The location of the fistula relative to the gastric tube was determined, and the 4 Fr catheter was positioned adjacent to the aortic wall of the fistula. The 4 Fr catheter tip was fixed to the fistula, and a 1.7 Fr microcatheter (Excelsior SL-10, Stryker Neurovascular, Fremont, California, USA) over a 0.014-inch guide wire (Transend EX, Stryker Neurovascular) was successfully inserted into the fistula using a coaxial technique (figure 2B). A small

Figure 1 Emergency contrast-enhanced CT in the arterial phase showing extravasation (white arrowhead) into the gastric tube through a thinner fistula (white arrow) from the descending thoracic aorta.

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Figure 2 Emergency abdominal aortography revealed remarkable extravasation of contrast medium that formed a pseudoaneurysm (black arrow) and flowed out of the gastric tube (black arrowheads) (A). A 1.7 Fr microcatheter passed through a 4 Fr catheter was inserted into the thinner fistula (arrow) with the coaxial technique. A small amount of contrast agent was injected into the fistula, including the pseudoaneurysm and extravasation (arrowheads). The fistula was then embolised with *n*-butyl cyanoacrylate/iodised oil through the microcatheter (B). Aortography immediately after embolisation (arrowheads) confirmed the complete absence of pseudoaneurysm and extravasation (C). Aortography after stent graft placement around the embolisation site (arrowheads) revealed persistent haemostasis (D).

amount of contrast agent was injected to confirm the position of the microcatheter relative to the fistula, and the volume of NBCA (Histoacryl; B Braun, Melsungen, Germany) mixed with iodised oil (Lipiodol Ultra Fluid, Guerbet, Aulnay-sous-Bois, France) required for embolisation was determined. The fistula and pseudoaneurysm were embolised by injecting 0.2 mL of NBCA and 0.2 mL of iodised oil through the microcatheter to enable fluoroscopic visualisation. The procedure was performed under digital subtraction angiography, and was stopped as soon as regurgitation of NBCA/iodised oil was observed (figure 2C). Aortography after embolisation confirmed the immediate and complete absence of extravasation from the fistula (figure 2D). The total duration of the procedure was 10 min, and the patient's circulatory dynamics stabilised immediately following the procedure.

A stent graft was placed under general anaesthesia for persistent haemostasis after the 5 h long TAE procedure. Three aortic stent grafts (Excluder Aortic Extender, W L Gore and

Associates, Flagstaff, Arizona, USA), with a maximum diameter of 28.5 mm and a length of 3.3 cm each, were partially overlapped and sutured around the NBCA embolisation site (figure 2D). Severe bilateral stenoses of external iliac arteries required the use of abdominal aortic stent grafts with a smaller diameter sheath compared to thoracic aortic stent grafts.

Although the patient was presented with the option of curative resection by replacing the aorta and gastric tube, she declined.

OUTCOME AND FOLLOW-UP

Contrast-enhanced CT at 21 days after embolisation showed no evidence of contrast extravasation. A high-attenuation area, corresponding to the fistula, and accumulation of NBCA/iodised oil were observed (figure 3). Follow-up endoscopy while the patient was taking a therapeutic proton pump inhibitor (PPI) revealed a reduction in peptic ulcer size. The patient was discharged 2 months postoperatively, with no clinical

Figure 3 Contrast-enhanced CT after embolisation showed no evidence of extravasation. Accumulation of *n*-butyl cyanoacrylate/iodised oil was observed, corresponding to the aortogastric tube fistula (white arrow). Aortic lesions were completely excluded, with no signs of endoleak or mediastinal infection/abscess.

complications. At 12 months postoperatively, contrast-enhanced CT revealed no extravasation and no other complications (eg, mediastinal abscess and ischaemia/infarction).

DISCUSSION

Aortic injury is a life-threatening condition with a high mortality rate, and thus requires rapid diagnosis and treatment. Small aortic injuries with arterial haemorrhage require an immediate response to restore haemostasis, and emergent arterial embolisation is one option.^{4–7} However, catheterisation can be difficult in these cases, because small target injuries (ie, fistula) branch out from larger arteries (ie, aorta). In this case, visualising the location of the fistula via non-invasive, contrast-enhanced CT improved the chances of a favourable outcome following microcatheter insertion into the aortogastric tube fistula.

Only a few reports have assessed the feasibility and efficacy of endovascular embolisation with NBCA/iodised oil for treating emergent haemorrhagic lesions.^{4–6} NBCA is a permanent liquid embolic material, and its adhesion time can be adjusted by mixing with iodised oil. A distinct advantage of NBCA/iodised oil is the dense radiopacity of the mixture, which allows observation of the exact site of occlusion during the embolisation procedure.⁸ Another advantage is that NBCA undergoes rapid polymerisation. Only a small amount of NBCA/iodised oil was needed to achieve haemostasis in our patient. We believe that our patient, who had critical haemorrhagic shock, received relatively safe embolisation under local anaesthesia, as compared to stent graft placement. Several reports have described the efficacy of embolisation using a combination of metallic coils and NBCA/iodised oil for thoracic aortic extravasation and/or pseudoaneurysm.⁷ In the present case, embolisation with coils was not an option given the fistula and extravascular space. However, embolisation with NBCA/iodised oil alone extended the amount of time available for considering and planning endovascular repair.

Common causes of this type of injury are anastomotic leakage and peptic ulcers. Several studies have reported the interval between oesophagogastric anastomosis and haematemesis in patients with anastomotic leakage to range from 11 to 80 days. This interval is shorter than that for patients who develop peptic ulcers, for whom the interval ranges from 14 months to 7 years.¹ The specific case reported here occurred 14 years after oesophagogastric anastomosis. The aetiology of peptic ulcer formation includes destruction of the mucosal barrier from ischaemia in the gastric tube and hypersecretion of gastric acid. Postoperative radiation and *Helicobacter pylori* infections are other possible causes of peptic ulcer formation; however, these factors were not found in our patient. The peptic ulcer may have resulted from psychological stress, which increases pulsation of the thoracic aorta and hypersecretion of gastric acid following discontinuation of PPI medications. PPI medications should be administered when evidence of excess gastric acid secretion exists. There are no specific signs that indicate the formation of an aortogastric tube fistula after intrathoracic oesophagogastric anastomosis. The only indications of this condition are vague symptoms, such as chest pain and heartburn. Once bleeding begins, it is very difficult to save the patient. The best course of treatment includes preventive care and careful, long-term follow-up of at-risk patients.

Emergent surgery for aortic rupture is challenging, and is associated with a high mortality rate.^{1–3} The recent availability of endovascular stent grafts has made it possible to treat aortic emergencies.^{2–3} However, endovascular management of aortogastric fistulas is associated with a high rate of recurrent bleeding and bacterial sepsis; additional radical surgery would therefore be necessary for long-term patient survival.^{9–11} The reason that none of these serious complications occurred in this case may be that the NBCA/iodised oil was injected between the stent graft and gastric tube. Stent graft repair was considered the first feasible option for interventional management for our patient. However, the patient was in critical haemorrhagic shock and immediate haemostasis was required.

One limitation of this report is that precise catheter placement may depend, in part, on operator skill. When performed by a practitioner with adequate experience, a high rate of successful aortogastric tube fistula embolisation using a microcatheter might be possible. Moreover, we did not consider the use of other embolic agents or particles that are effective for treating haemorrhage.

In conclusion, this report details a very rare case of postoesophagectomy aortogastric tube fistula with massive bleeding. Visualisation and accurate targeting of the aortic injury site, as well as selective fistula embolisation with NBCA/iodised oil, proved effective for treating the initial extravasation, particularly for a substantially thinner aortogastric tube fistula. After initial embolisation, additional stent graft placement may achieve higher haemostasis efficacy.

Learning points

- ▶ Aortogastric tube fistula with massive gastrointestinal bleeding is a very rare condition; however, there is a possibility that this complication arises after transhiatal subtotal oesophagectomy.
- ▶ Aortogastric tube fistula in the present case was likely due to peptic ulcer formation.
- ▶ Use of *n*-butyl cyanoacrylate/iodised oil embolisation could provide an effective and immediate treatment option for aortogastric tube fistula with critical haemorrhagic shock.

Competing interests None.

Patient consent Obtained.

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REFERENCES

- 1 Takebayashi T, Okushiba S, Ohno K, *et al.* Peptic ulcer induced acute aortogastric fistula occurring 7 years after a pharyngogastrostomy following a resection for carcinoma of the oesophagus: report of a case. *Surg Today* 2004;34:777–9.
- 2 Sato O, Miyata T, Matsubara T, *et al.* Successful surgical treatment of aortogastric fistula after an oesophagectomy and subsequent endovascular graft placement: report of a case. *Surg Today* 1999;29:431–4.
- 3 Takano S, Katsuhara K, Nobuhara K, *et al.* Aorto-esophageal fistula due to esophageal ulcer. *Gen Thorac Cardiovasc Surg* 2009;57:255–7.
- 4 Kawai N, Sato M, Tanihata H, *et al.* Repair of traumatic abdominal aortic pseudoaneurysm using N-butyl-2-cyano-acrylate embolization. *Cardiovasc Intervent Radiol* 2010;33:406–9.
- 5 Cantasdemir M, Adaletli I, Cebi D, *et al.* Emergency endovascular embolization of traumatic intrarenal arterial pseudoaneurysms with N-butyl cyanoacrylate. *Clin Radiol* 2003;58:560–5.
- 6 Monsignore LM, Scarpelini S, Santos JS, *et al.* Urgent percutaneous transcatheter embolization of hemorrhagic hepatic lesions with N-butyl cyanoacrylate. *Diagn Interv Radiol* 2012;18:403–9.
- 7 Sanchez MJ, Ananian CL, Berkmen T. Embolization of an aortic arch pseudoaneurysm with coils and N-butyl-cyanoacrylate. *J Vasc Interv Radiol* 2006;17:1677–9.
- 8 Koganemaru M, Abe T, Hayabuchi N, *et al.* Successful endovascular treatment of iatrogenic pseudoaneurysm with a relatively long neck. *BMJ Case Rep* 2013;2013: pii: bcr2012007021.
- 9 Kakkos SK, Papadoulas S, Tsolakis IA. Endovascular management of arterioenteric fistulas: a systemic review and meta-analysis of the literature. *J Endovasc Ther* 2011;18:66–77.
- 10 Canaud L, Ozdemir BA, Bee WW, *et al.* Thoracic endovascular aortic repair in management of aorto-esophageal fistulas. *J Vasc Surg* 2014;59:248–54.
- 11 Flores J, Shiiya N, Kuniyama T, *et al.* Aorto-esophageal fistula: alternatives of treatment case report and literature review. *Ann Thorac Cardiovasc Surg* 2004;10:241–6.

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