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INTERVENTIONAL RADIOLOGY
CASE REPORT

Occlusion of a neo-esophageal-bronchial fistula using the Amplatzer Vascular Plug 2

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ABSTRACT
The appearance of a benign fistula between the airway and the gastrointestinal tract is a rare complication of esophagectomy. We report a patient with neo-esophago-bronchial fistula that developed 13 months after two-stage esophagectomy. Repeat thoracotomy was not deemed appropriate given the patient's chronic sepsis and malnutrition. After unsuccessful attempts at endoscopic closure, the fistula was successfully and permanently occluded under radiological guidance with an Amplatzer® Vascular Plug 2. The patient remained asymptomatic, with a measured weight gain, 12 months after the successful fistula occlusion.

Benign bronchial fistulae are uncommon after esophagectomy. The clinical presentation varies from minimal symptoms, such as cough, to sepsis and mediastinitis (1). The rarity of the complication, combined with its often nonspecific symptoms and the poor sensitivity of computed tomography (CT) and endoscopy for its detection, makes the diagnosis difficult (2). Ulceration of the gastric tube, or neo-esophagus, is thought to be the source of late fistulation. Several mechanisms have been suggested for this phenomenon, including inadequate pyloric gastric tube drainage, breakdown of esophageal mucus and the mucosal barrier due to vascular disturbances, the effects of postoperative irradiation, and peptic ulceration (1).The treatment depends on a variety of factors including the patient's symptoms, current health status, comorbidities, and the position and size of the fistula. Well-documented treatment modalities include surgery, stenting, tissue glue, coagulation and clipping; treatments may be used either singly or in combination. Here, we report a patient with neo-esophago-bronchial fistula that developed 13 months after two-stage esophagectomy and successfully occluded with an Amplatzer® Vascular Plug 2 (AVP 2; AGA Medical Corp., Plymouth, Minnesota, USA).

Case report

A 71-year-old woman was diagnosed by endoscopy with a squamous cell carcinoma in the mid-esophagus. The lesion was staged as T3N0M0; following neoadjuvant chemotherapy with cisplatin/5-fluorouracil, the patient underwent a two-stage esophagectomy. In brief, this involved a laparotomy with gastric mobilisation, left gastric-territory lymphadenectomy, and formation of a gastric tube to make a neo-esophagus. The abdomen was closed, the patient moved to the right lateral position, and thoracotomy was performed. Esophageal resection with lymphadenectomy was carried out, followed by reconstruction via the posterior mediastinal route. A CEEA 25 stapler (Covidien, Mansfield, Massachusetts, USA) was used to make an end-to-side stapled esophago-gastric anastomosis and two chest drains were placed. The patient's postoperative recovery was uneventful, apart from a single episode of medically treated atrial fibrillation on postoperative second day. The apical chest drain was removed on day four and, following a satisfactory water-soluble contrast swallow, the basal chest drain was removed on day six. Pathologic examination of the esophagectomy specimen showed a complete response to chemotherapy with no residual cancer cells detectable. The patient followed up regularly and had no complaints, although it was noted that her weight gain was poor.

Thirteen months after the curative resection she presented to the emergency department with shortness of breath and was treated for community-acquired pneumonia; this occurred twice over a two-month period.

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During her follow-up in the respiratory and surgical clinics, she described the new symptom of a persistent cough at night and after eating. CT and a water-soluble contrast swallow with video-fluoroscopy were performed, looking for evidence of either aspiration or recurrence of her cancer. The only pertinent finding was on CT: a solitary enlarged mediastinal lymph node. Her symptoms remained unchanged for the next 18 months, at which time she experienced a right-sided pneumonia. CT was repeated and esophago-gastro-duodenoscopy was performed, with no new findings to explain her symptoms. The patient was followed up in the respiratory clinic and was noted to have right basal crackles and poor weight gain; she went on to have another episode of pneumonia. At that time, endoscopy and a barium swallow examination were performed. The barium swallow demonstrated a fistula between the gastric tube forming the neo-esophagus and the right upper-lobe bronchus (Fig. 1). A naso-jejunal tube was inserted endoscopically for nutrition. An endoscopic attempt to close the fistula by glue obliteration and clipping was unsuccessful as the fistula could not be cannulated.

An attempt at fistula closure under radiological guidance was then planned. The patient was placed in the prone position, and fluoroscopic guidance was used to gain oral access to the upper neo-esophagus. Contrast was injected to delineate the fistulous tract and a hydrophilic guidewire (0.035 inches regular angled Terumo Glidewire, Terumo Medical Corporation, Somerset, New Jersey, USA) was passed through the fistula into the right upper-lobe bronchus (Fig. 2). A 7 F, 55 cm Terumo Destination vascular sheath (Terumo Medical Corporation) was then introduced over the guidewire, and a 14 mm AVP 2 was deployed. The distal end of the plug was positioned in the fistula tract without entering the bronchus, and remained slightly elongated. The proximal end was in the neo-esophagus and was flush with the orifice of the fistula, ensuring that the orifice was closed. A water-soluble contrast swallow examination was performed 48 hours later demonstrating complete occlusion of the fistula (Fig. 3). One year after this procedure, the patient has remained asymptomatic and has achieved a measured weight

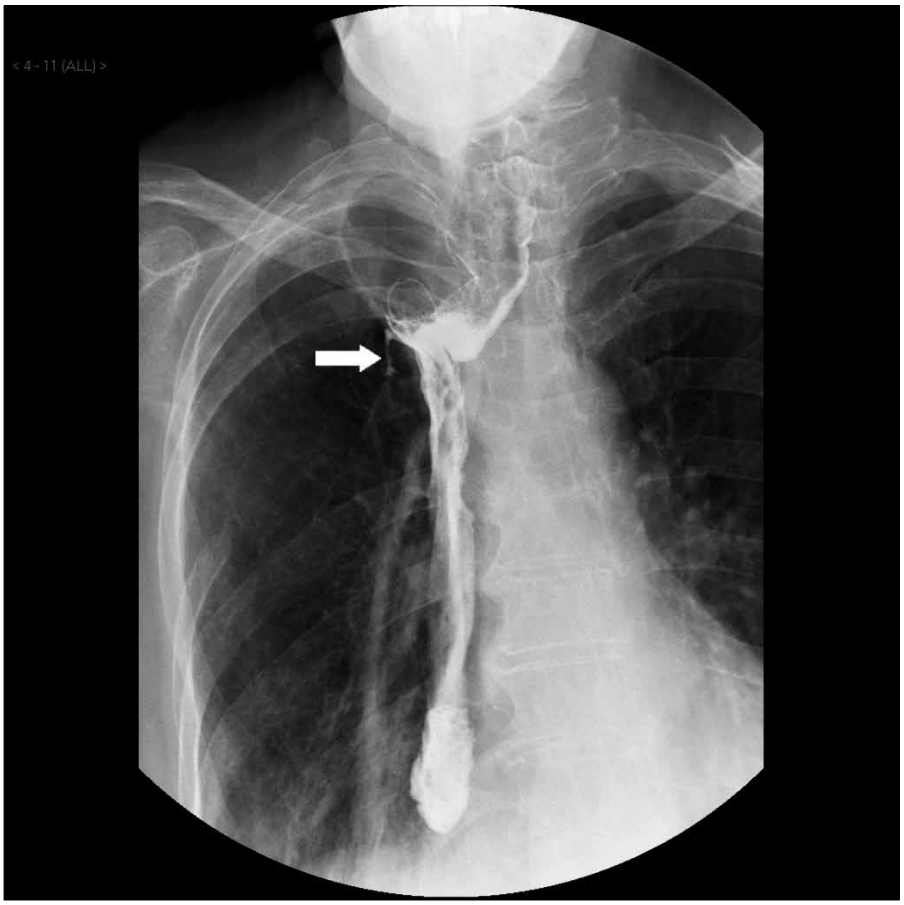

Figure 1. Oral contrast leak from the gastric tube consistent with neo-esophago-bronchial fistula (*arrow*).

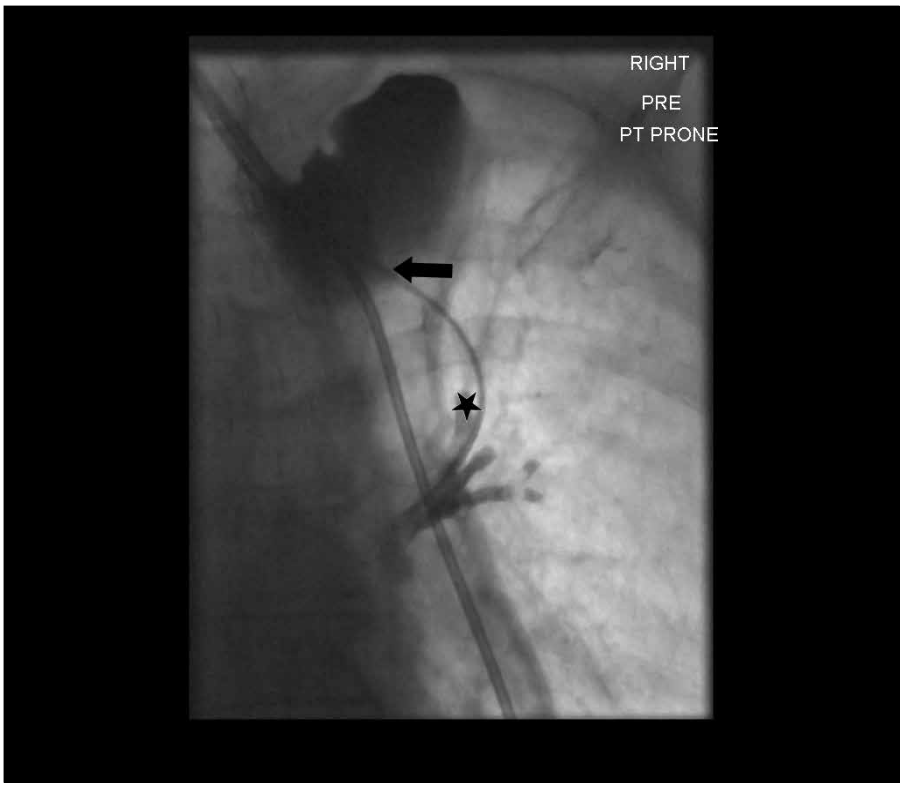

Figure 2. Contrast injection through the fistula (*arrow*) demonstrates its connection to the segmental bronchus (*star*).

gain. The device may be seen in position on chest radiography (Fig. 4).

Discussion

The diagnosis of a neo-esophago-bronchial fistula can often be difficult and may take a number of years. Barium esophagography is regarded as more sensitive than endoscopy (esophago-gastro-duodenoscopy or bronchoscopy) or CT in the diagnosis of this condition (2). Once diagnosed, the procedure for surgical repair of an esophago-bronchial fistula is well documented: excision of the fistula tract, removal of permanently damaged lung tissue, and interposition of viable tissue (1). The mortality of this procedure, however, is 10% with up to 50% morbidity. Repeat thoracotomy performed in chronically malnourished and septic patients may explain these high rates. Stenting is a well-documented treatment for malignant fistulae, however the lack of stricture predisposes to migration of the stent when stenting is used for benign fistulae (2).

Endoscopic treatment is another recognised, and now well-established option for fistula treatment. Tissue glue and clipping may be used, or a combination may be employed of glue and clipping or argon electrosurgical ablation and clipping. Endoscopic treatment with tissue glue is cheap and is performed under direct visualisation, but it has a high failure rate, particularly when the fistula tract is short or wide, as the fibrin glue can be washed out. Endoscopic clipping can be difficult, especially when the fistula tract is mature. A combination of glue and clipping may improve the chances of success, although repeated attempts may be necessary (2). Endoscopic attempts at treating the fistula in this patient failed due to inability to cannulate the fistula for glue placement and to achieve the proper position for clip placement that would occlude the fistula lumen. These attempts failed because of a protrusion of the fistula tract into the bronchus, making cannulation and device placement challenging. A stent was deemed likely to migrate and was therefore not attempted. The next step was closure under radiological guidance. The prone position was chosen on the basis of the fistula tract protrusion seen at endoscopy, and on the clinical grounds that gastric tube protrusion is asymptom-

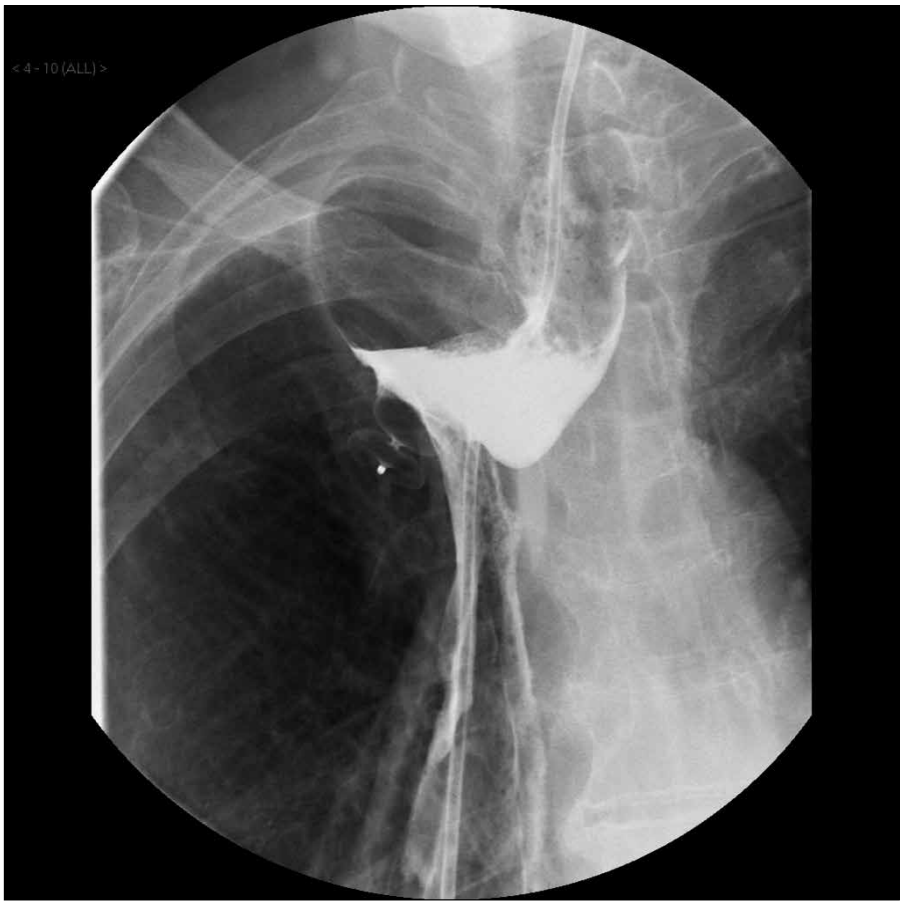

Figure 3. Water-soluble contrast swallow on day two shows the plug deployed with the proximal end within the neo-esophagus and the distal end remaining within the fistula tract rather than entering the bronchus. The contrast is contained within the gastric tube.

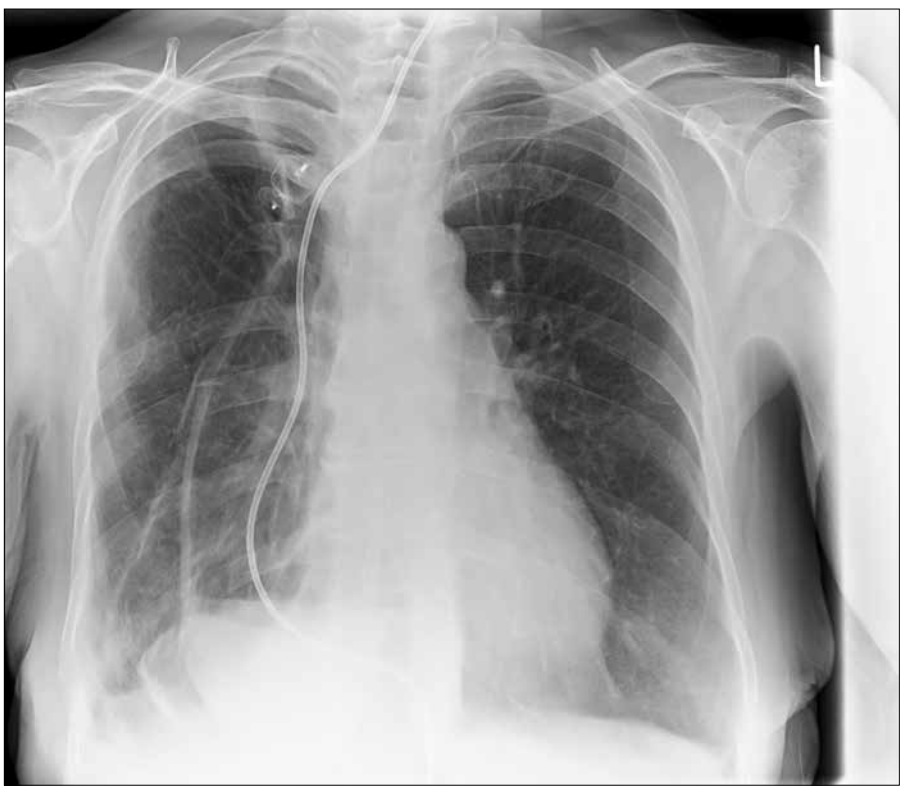

Figure 4. Chest radiography shows the plug in situ.

atic whereas protrusion into the true bronchus often results in irritation and cough. Radiologically-guided methods for fistula closure include coils, tissue glue (n-butyl 2 cyanoacrylate), and purpose-designed vascular devices. The use of glue is rapid and the material can be delivered through microcatheters due to its low viscosity, but its use is associated with proximal and distal migration, catheter entrapment, and recanalisation of the fistula. The use of single coils is cheap, but multiple coils are often required in this situation. Coils cause significant CT artifact and carry the risk of incomplete occlusion and of distal migration or embolism. Occluding devices may be more expensive than single coils, but they are cheaper when the use of multiple items is taken into account. The advantages of these devices are their rapid deployment, their small-diameter catheter delivery systems, and their precisely targeted placement, with repositioning or removal easily achieved; device migration may still occur, however (3, 4). Amplatzer devices are permanent vascular occlusion devices made of ninitol mesh and are designed to cause vessel thrombosis. There are various models (1, 2, 3, and 4) with varying shapes and sizes. The AVP 2 has dual "lozenges" of polyester fabric rather than the single disc of the AVP 1. Its advantages include its availability in a range of sizes, an instant mechanical seal, and the generation of long-term tissue granulation to reinforce the seal. Its use is well-described for the permanent closure of atrial septal defects and in the closure of medium-sized arteries (4). The ninitol mesh has a memory and tries to return to its normal shape when deployed. When it is used in vessels, an oversize of 30%–50% is recommended as there is a slightly variable but predictable rate of elongation (3). Sizing for vessel occlusion can be measured using standard fluoroscopic techniques. The off-licence use of this device in non-anatomical fistulous tracts is not common. It has recently been used in a small number of esophageal fistulae, utilizing the concept that granulation tissue will eventually occlude the fistula. It has been visually placed during bronchoscopy (5–7) or placed under radiologic guidance into a cavity after packing the cavity with coils and glue (8). This is the first description of using an isolated AVP 2 to close a neo-esophago-bronchial fistula via the gastrointestinal tract under solely radiological guidance.

In conclusion, benign neo-esophageal-bronchial fistulae are a rare cause of recurrent pneumonia and can be difficult to diagnose. Endoscopic management with tissue glue and clips is often the first-line of treatment, but the placement of vascular plugs under radiologic guidance is a viable alternative. This technique avoids the need for repeat thoracotomy with its associated morbidity and mortality.

Conflict of interest disclosure

The authors declared no conflicts of interest.

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