



Code Stroke Postendoscopy: A Case of Pneumocephalus From a Spinal-Esophageal Fistula

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ABSTRACT

Esophageal fistula to the respiratory tract and mediastinum is a well-described complication from esophageal malignancies. Spinal-esophageal fistula (SEF) on the other hand is a much rarer complication that has only been reported in few instances. Here, we report a unique case of fatal spinal-esophageal fistula with an associated pneumocephalus in an 83-year-old woman with metastatic esophageal squamous cell carcinoma.

KEYWORDS: spinal-esophageal fistula; esophageal squamous cell carcinoma; stent; pneumocephalus

INTRODUCTION

A spinal-esophageal fistula (SEF) is an extremely uncommon type of fistula between esophagus and spinal tract that can lead to serious complications, such as pneumocephalus, encephalitis, paraplegia, central nervous system infection, and death. SEF has previously been described in rare cases of anterior cervical surgery, discitis, and esophageal malignancy postradiotherapy.¹⁻³ However, SEF in patients with esophageal malignancy without prior radiotherapy or forementioned risk factors has not been reported. We present a unique case of SEF complicated by pneumocephalus in a patient with esophageal squamous cell carcinoma without a history of discitis, cervical spine surgery, or radiotherapy presenting with stroke-like deficits post-esophageal stenting.

CASE REPORT

An 83-year-old woman with hypertension and atrial fibrillation presented with a 3-month history of progressive dysphagia to solids. An esophagogastroduodenoscopy (EGD) was performed revealing an obstructive circumferential esophageal mass at 21 cm from the incisors extending distally over 7 cm. Biopsies of the mass confirmed esophageal squamous cell carcinoma (SCC). Full-body computed tomography (CT) and positron emission tomography showed surrounding enlarged lymph nodes but no distant metastases. Her case was reviewed by a multidisciplinary team including thoracic surgery and oncology. She was deemed unsuitable for surgical intervention because of the presence of lymph node involvement and was planned for conformal radiation therapy. While awaiting therapy, her dysphagia progressed to both solids and liquids; therefore, endoscopy was repeated and a fully covered 18 mm × 10 cm esophageal stent was placed. Subsequently, her dysphagia improved significantly and was discharged home with plans for outpatient radiotherapy.

She was seen in follow-up 6 weeks later for her radiotherapy appointment but was found to be unwell with chest pain and in atrial fibrillation with a rapid ventricular rate, along with worsening dysphagia to both solids and liquids. She was subsequently admitted to hospital and managed supportively with intravenous hydration. Chest CT demonstrated interval progression of the esophageal mass with tumor overgrowth at the distal edge of the stent causing obstruction. A new focal nodularity within the left upper lung lobe had also developed suggesting metastases. Given the CT findings and her dysphagia symptoms, an EGD using carbon dioxide insufflation was performed to evaluate for stent dysfunction. This confirmed progression of her esophageal SCC

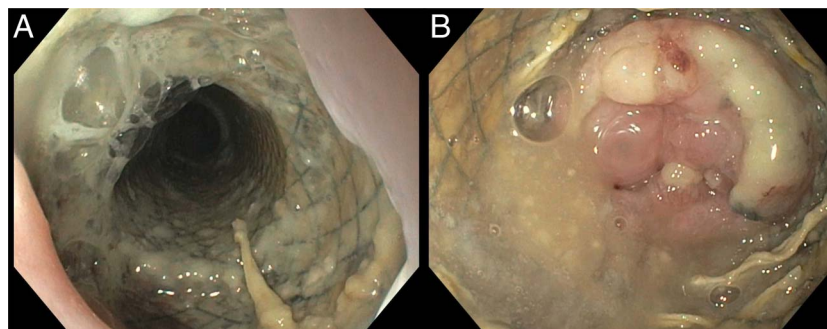


Figure 1. Esophagogastroduodenoscopy showing esophageal stent in situ (A) and tumor overgrowth distally (B).

with tumor extension into the distal aspect of her metal stent (Figure 1). Proximal to the stent and her previously known malignancy, there was a lobulated and protruding focal area concerning for infiltration and further extension of her SCC (Figure 2). A large defect was appreciated at the proximal end of the mass, and fistulization or impending fistulization was suspected. As such, the esophageal stent was replaced with a longer 18 mm × 15 cm fully covered metallic stent placed over the defect (Figure 3).

Several hours after EGD, she had a sudden clinical deterioration. She became hypotensive with a decreased level of consciousness. She had associated anisocoria with a pinpoint left pupil and left facial droop. A code stroke was activated, and the neurology service was consulted. Urgent CT of the head and neck did not show acute cerebral infarct or intracranial hemorrhage, but the esophageal soft-tissue mass appeared to be contiguous with paraspinal soft tissue at the level of T1-T2. In addition, pockets of gas were seen within the right paraspinal soft tissue, extending through the right T1-T2 neural foramen, along with gas locules in the cervical-thoracic spinal canal (Figure 4), suprasellar and prepontine

cisterns consistent with a SEF with associated pneumocephalus (Figure 5). When previous cross-sectional imaging was reviewed, there was a small amount of air noted within the spinal canal suggesting the fistula likely preceded the endoscopic procedure (Figure 6). Given her extensive malignancy and frailty, her condition was deemed irreversible. She was then transitioned to palliative care and subsequently died on the following day.

DISCUSSION

Several types of esophageal fistula have been described in the literature. Esophageal-respiratory (i.e., tracheoesophageal and bronchoesophageal), aorto-esophageal, and esophago-mediastinal fistulas are the most reported types. Esophageal fistula to spinal tract is very rare and reported only in settings of radiation therapy, diskitis, and postoperative complication from anterior cervical spine surgery and recently from a severe esophageal ulcer.^{1,3-5} Although esophageal fistula formation is a known complication of esophageal self-expanding metallic stent, fistulas reported include aorto-esophageal fistula or esophageal-respiratory

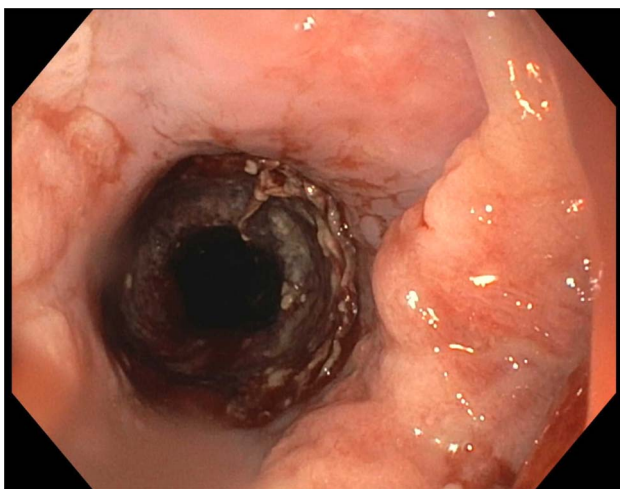


Figure 2. Esophagogastroduodenoscopy showing a focal protruding area proximal to the location of the previously visualized squamous cell carcinoma concerning for an infiltrative and potential fistulizing process.

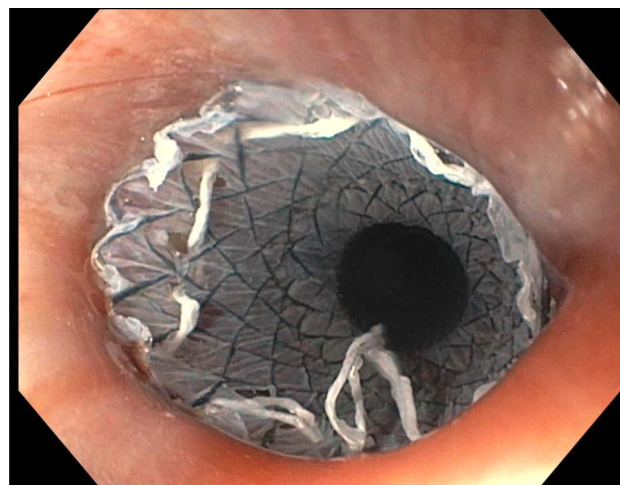


Figure 3. Esophagogastroduodenoscopy after placement of an 18 mm × 15 cm fully covered metallic stent over the concerning defect.



Figure 4. Head and neck computed tomography demonstrating gas locules (arrow) in the cervical-thoracic spinal canal.

fistula. There has been no report of SEF secondary to stenting to the best of our knowledge.^{6,7} Our case was also likely acquired secondary to esophageal malignancy rather than stenting, but it may have been exacerbated with new



Figure 5. Head computed tomography demonstrating gas locules (arrow) in the suprasellar and prepontine cisterns consistent with pneumocephalus.

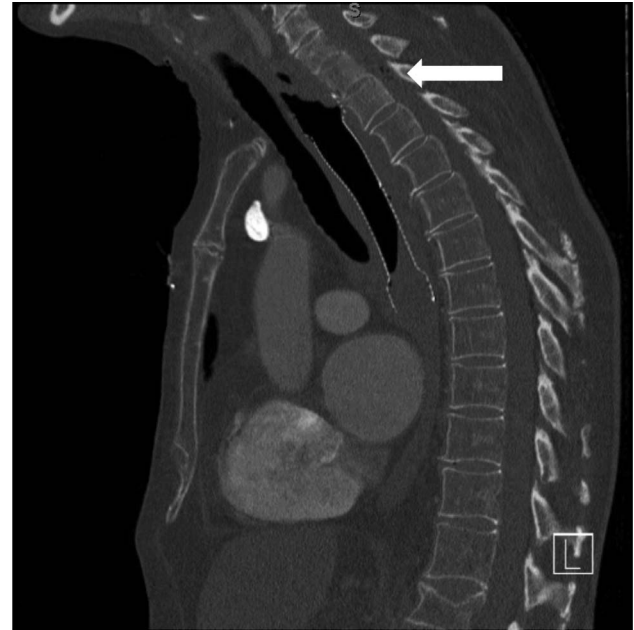


Figure 6. Chest computed tomography showing the presence of small gas locules within the spinal canal (arrow) before stent exchange.

pneumocephalus by endoscopic stenting given clinical timeline.

On our review of the literature, this is the first reported case of SEF with associated pneumocephalus secondary to esophageal malignancy without prior radiotherapy or cervical spine surgery. In addition, this case is unique because the patient suffered a fatal complication with pneumocephalus postesophageal stenting which has not been reported secondary to SEF.⁸

DISCLOSURES

Author contributions: A. Fetz: patient evaluation and management and drafting the manuscript. F. Ghaseminejad: drafting the manuscript. H. Kim: critical revision of the manuscript. S. I. Gan: patient evaluation and management, critical revision, and is the article guarantor.

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Informed consent was obtained for this case report.

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