Successful Airway Stenting Using Silicone Prosthesis for Esophagobronchial Fistula*

Keisuke Miwa, MD; Masahiro Mitsunaga, MD, FCCP; Kohsuke Tayama, MD; Naofumi Tomita, MD; Shinzo Takamori, MD, FCCP; Akihiro Hayashi, MD; and Kazuo Shirouzu, MD

We present the case of a 55-year-old man with advanced esophageal cancer who was successfully treated using a self-expandable metallic stent (S-EMS) for 6 months and subsequently was treated for an esophagobronchial fistula as a complication of the initial S-EMS using a silicone airway stent for an additional 4 months. This is the first report in the literature concerning penetration into the airway of an S-EMS implanted in the esophagus. The present case suggests that airway stenting using a silicone stent as treatment for an esophagobronchial fistula may represent a useful modality.

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Key words: bronchial fistula; esophageal fistula; self-expandable metallic stent; silicones; stent–related complication

Abbreviation: S-EMS = self-expandable metallic stent

Stent therapy using a self-expandable metallic stent (S-EMS) in patients with esophageal stenosis has resulted in improvements to the quality of life for patients with inoperable esophageal cancer.1–3 However, stent-related complications such as hemorrhage, rupture, stent migration, granulation tissue formation, and esophagotracheobronchial fistula have been reported.4 In particular, the management of esophagotracheobronchial fistulas presents difficulties. We report a case of an esophagobronchial fistula due to penetration of an S-EMS that had been implanted in the esophagus, which was successfully treated by silicone stent placement in the bronchus.

Case Report

A 55-year-old man with dysphagia due to esophageal cancer was referred to our hospital. A barium esophagogram and CT scan revealed severe stenosis in the middle esophagus and abdominal lymph node metastasis. Squamous cell carcinoma was diagnosed by endoscopic biopsy and was clinically staged as T4N3M0 stage IV on the basis of imaging by CT scan and esophagography. The patient initially received chemoradiotherapy.

Tumor-specific chemotherapy (with cisplatin and fluorouracil) and radiotherapy were undertaken. The chemotherapy regimen consisted of IV administration of 100 mg cisplatin on day 1 and 1,000 mg fluorouracil on days 1 to 4. This course of treatment was administered three times every 3 weeks. During this treatment, a 3.0-Gy dose fraction of radiation per day, 5 days per week, was administered for 4 weeks, for a total dose of 60 Gy. The radiation field included the primary tumor (with a 2.0-cm margin of healthy esophageal tissue), the infraclavicular trachea, and the left main bronchus.

As a result, although the tumor was reduced to 56% of its original size, the esophageal lumen remained stenosed and the enlarged abdominal lymph nodes displayed almost no change compared to the condition before chemoradiotherapy. Stent therapy was subsequently selected to reduce esophageal stenosis. A covered S-EMS (Covered Ultraflex Esophageal Stent System, Microinvasive; Boston Scientific Co; Boston, MA) was placed in the esophagus on the 10th day after chemoradiotherapy. The course after stent placement was initially uneventful. However, the patient complained of severe cough on oral intake in the sixth month after stent placement. Esophagography revealed an esophagobronchial fistula, and an additional stent of the same type was introduced into the previous stent in the esophagus. Although initial improvement was observed, symptoms recurred within 1 month. Esophagography and CT scanning revealed an esophagobronchial fistula. Moreover, bronchoscopic findings revealed S-EMS penetration to left main bronchus (Fig 1). The patient subsequently received a silicone stent (Dumon Tube BD; Novatech Co; Aubagne, France) that was placed in the left main bronchus. A rigid bronchoscope was inserted under general anesthesia immediately before the second carina while slowly rotating and holding down the S-EMS, which penetrated into the left main bronchus through the vocal cords. A sheath containing the appropriate size silicone stent subsequently was inserted through the rigid bronchoscope, and the stent was released from the sheath using a pusher. The final stent position then was adjusted using forceps.

The symptoms of an esophagobronchial fistula subsequently resolved. Oral intake was resumed on the 29th day after silicone stent placement, as esophagography did not reveal an esophagobronchial fistula. Although patient recovery followed a satisfactory course, death due to cancer occurred in the fourth month after airway stent placement.
DISCUSSION

Although esophageal stent therapy is only a palliative measure for patients with inoperable tumors, it remains an important method for maintaining quality of life. In particular, the usefulness of a covered S-EMS as therapy for a malignant esophageal obstruction has been reported. Double stenting for both esophageal and tracheobronchial stenosis represents a useful technique. However, the indications for airway stenting in patients with inoperable, malignant, esophageal stenoses, but without airway stenoses, have become a matter of some contention. The timing of the esophageal stent insertion is closely associated with stent-related complications. Yorozu et al reported that esophageal stent placement within 1 month after the patient has undergone radiotherapy is associated with a high risk of rupture. In the present case, esophagobronchial fistula may have occurred because the insertion of an S-EMS closely followed radiotherapy. Reintervention with an S-EMS initially improved the symptoms of the fistula. However, we believe that an additional stent should not be inserted into previous stents in cases of esophagobronchial fistula. We suspect that an exposed part of the initial stent penetrated into the left main bronchus following the expansion of the second stent.

We selected the Dumon prosthesis as an airway stent to block the fistula. This choice was made because we thought that placement of an S-EMS in the left main bronchus might exacerbate the esophagobronchial fistula, and because an S-EMS that is made of nitinol for airway stenosis is not yet covered by the national health insurance system in Japan. Some reports already have addressed the utility of silicone stents in patients with malignant airway stenoses. This type of stent is covered by the national health insurance system in Japan. In the present case, the silicone stent appeared to block the fistula and improved the oral alimentation status of the patient.

CONCLUSION

The present case suggests that airway stenting using a silicone stent, instead of an esophageal stent, for the treatment of an esophagobronchial fistula may represent a useful modality.

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Pulmonary Embolism in a Patient With Pernicious Anemia and Hyperhomocysteinemia*

Angel Caldera, MD; Jorge Mora, MD; Morris Kotler, MD; and Glenn Eiger, MD

We report the case of a 60-year-old woman with a history of ataxia who sought evaluation after a syncopal episode. A diagnostic workup revealed pulmonary emboli, pernicious anemia (PA), hyperhomocysteinemia, and a G20210A prothrombin gene mutation. She was successfully treated with homocysteine-lowering therapy, including high doses of oral cobalamin. She also received oral anticoagulation for 6 months. At 1 year of follow-up, no further thrombotic episodes had occurred. Our report highlights the thrombogenic risk of hyperhomocysteinemia secondary to PA in a patient with the G20210A prothrombin gene mutation.

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Key words: cobalamin; hyperhomocysteinemia; pernicious anemia; prothrombin gene variant; pulmonary embolism; thrombophilia

Abbreviations: IF = intrinsic factor; MMA = methylmalonic acid; PA = pernicious anemia

Pernicious anemia (PA) is commonly associated with hyperhomocysteinemia.1 The purpose of this report was to highlight the thrombogenic risk of hyperhomocysteinemia secondary to PA. Additionally, our patient was found to be a carrier of the prothrombin gene mutation. To the best of our knowledge, this is the first report of pulmonary thromboembolic disease that possibly was triggered by hyperhomocysteinemia secondary to PA.

CASE REPORT

A 60-year-old African-American woman sought evaluation after a syncopal episode. She complained of an unsteady gait for 3 weeks prior to hospital admission but denied dyspnea, chest pain, or dizziness. Her medical history was remarkable for well-controlled hypertension. There was no history of thrombosis, miscarriages, recent physical trauma, or prolonged air travel. The initial assessment revealed an ill-appearing woman with a BP of 110/75 mm Hg, heart rate of 130 beats/min, and respiratory rate of 25 breaths/min. Pulse oximetry was 94% on room air. A loud pulmonic component of the second heart sound and non-cerebellar ataxia were present. The results of the rest of the physical examination were within normal limits. Laboratory data showed an increased mean corpuscular volume and a normal hemoglobin level. Biochemical and clotting profiles were normal. The ECG showed sinus tachycardia without right heart strain. Chest radiograph findings were normal. Arterial blood gas levels measured with the patients breathing room air revealed the following: pH, 7.46; PaCO₂, 25 mm Hg; and PaO₂, 73 mm Hg. Lower extremity Doppler scans were negative for deep venous thrombosis. The results of a ventilation/perfusion lung scan were abnormal (Fig 1, middle, A). The patient received a diagnosis of pulmonary embolism and began therapy with IV heparin. Further extensive laboratory data are presented in Table 1. She was discharged from the hospital while receiving therapy with homocysteine-lowering agents, including folate (1 mg/d), pyridoxine (100 mg/d), and high-dose oral cobalamin (1,500 μg/d). She continued to receive oral anticoagulation therapy for 6 months. Serum methylmalonic acid (MMA), homocysteine, and cobalamin levels normalized after 3 months of therapy (Table 1). A repeat ventilation/perfusion lung scan 6 months later demonstrated significant improvement (Fig 1, bottom, B). She recovered completely from her ataxia. After 1 year of follow-up, the results of the patient’s age-appropriate cancer screening examination has been negative. There have been no further episodes of thrombosis.

DISCUSSION

PA is the most common cause of cobalamin deficiency. Intrinsic factor (IF), a glycoprotein that is produced by the gastric parietal cells, is essential in facilitating cobalamin absorption. An autoimmune insult against these cells causing decreased IF production is the usual basis for this disorder.2 Since cobalamin is required in the folate-dependent remethylation of homocysteine to methionine and in the folate-independent conversion of methylmalonyl-coenzyme A to succinyl-coenzyme A, serum levels of homocysteine and MMA are increased in 95% of patients with cobalamin deficiency.1 Hyperhomocysteinemia is a well-recognized risk factor for thrombosis and atherosclerotic vascular disease.3

Currently, PA can be diagnosed in the early stages based on the presence of anti-IF/anti-parietal cell antibodies even before alteration of hematologic parameters. In a series of 100 patients with PA, 70% had hemoglobin levels of >12 g/dL, and 36% have a mean corpuscular volume of <100 fl, at the time of diagnosis.4 Peripheral polyneuropathy (ie, paresthesias and sensory ataxia) along with subacute combined spinal cord degeneration are well-recognized neurologic manifestations of the disease. If the initiation of cobalamin replacement therapy is delayed, the neurologic deficits may not revert.2 Given that about 2% of cobalamin intake is passively absorbed through an IF-independent pathway,5 we treated our patient with high doses of oral cobalamin, avoiding the parenteral route in the setting of anticoagulation. Her clinical recovery, evidenced by the reversal of neurologic symptoms, was likely due to the early diagnosis and successful treatment of the cobalamin deficiency.

Our patient also was found to be a carrier of the G20210A prothrombin gene mutation. This genetic defect has been described in 2.5% of healthy individuals6 and is...