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Case Report

Minimally Invasive Approach to Esophageal Perforation after Endoscopic Ultrasound-Guided Fine-Needle Aspiration: A Report of 2 Cases

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Esophageal perforation after endoscopic ultrasound-guided fine-needle aspiration for mediastinal staging is a rare but severe complication. We report 2 cases of patients with esophageal perforation who were treated using video-assisted thoracoscopic surgery in combination with esophageal stenting. Through these cases, the feasibility of minimally invasive thoracic surgery was evaluated.

Keywords: Endoscopic procedures, Video-assisted thoracic surgery, Injury, Esophageal perforation

Case report

Endoscopic ultrasound-guided fine-needle aspiration (EUS-FNA) has emerged as a novel minimally invasive imaging technique to obtain a histologic diagnosis in patients with mediastinal lymphadenopathies accompanying various benign or malignant conditions. It complements advanced imaging methods such as positron emission tomography-computed tomography (PET-CT), which have a high sensitivity for identifying enlarged lymph nodes, but limited specificity. In particular, differentiating between inflammatory processes and malignancy is especially difficult and a pathological diagnosis is generally required. The overall sensitivity and specificity of EUS-FNA and/or endobronchial ultrasound (EBUS)-transbronchial needle aspiration were reported to be 97.4% and 100%, respectively [1]. Although severe complications are rare, we report 2 serious and life-threatening cases of esophageal wall rupture after EUS-FNA. Surgery via thoracotomy to clean the thoracic cavity and close the esophageal defect is the gold standard treatment for esophageal perforations, but less invasive treatments are being explored and their role is evolving. Hence, we report 2 cases of esophageal wall rupture after EUS-FNA in which minimally invasive surgery was successfully combined with endoscopic stenting.

This study was conducted according to the principles of the Declaration of Helsinki. Verbal informed consent was obtained from both patients for publication of their clinical details and images.

Case 1

Patient A was a 45-year-old man who underwent a work-up for recurrent right-sided chest pain and increased D-dimer levels. Computed tomography (CT) was performed to exclude pulmonary embolism and showed mediastinal and hilar lymphadenopathy, which raised the suspicion of lymphoma or sarcoidosis. An additional PET-CT examination showed axillary and mediastinal enlarged PET-positive lymph nodes, most prominently in the subcarinal region. In addition, paraseptal emphysema and thickened peribronchial interstitium in the apex of the lung were present, with signs of active fibrosis. EUS-FNA of lymph node station 7 was performed with a 19G needle for patho-

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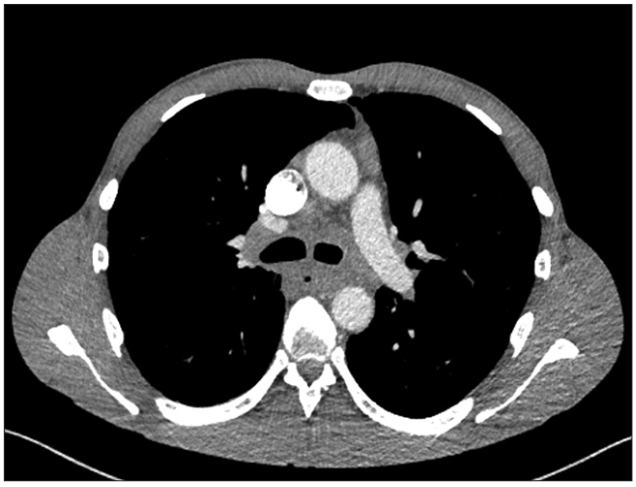

Fig. 1. Computed tomography scan showing mediastinitis.

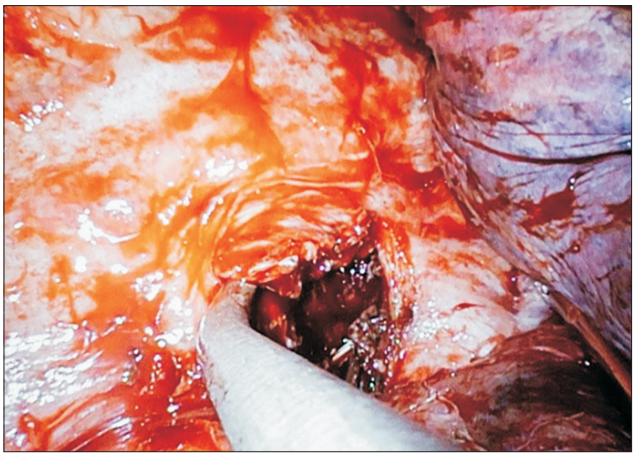

Fig. 2. Uniportal video-assisted thoracoscopic surgery showing a tear in the esophagus.

logical and microbiological evaluation.

The patient was readmitted after 12 days with excessive vomiting, followed by epigastric pain and fever. The patient was hemodynamically stable with a blood pressure of 130/80 mm Hg, a regular heart rate of 80 beats/min, a temperature of 37.2°C, and a respiratory rate of 15 breaths/min. His C-reactive protein (CRP) level was 243 mg/L and white blood cell (WBC) count was 18×10^9/L.

Chest CT showed posterior mediastinitis with possible abscess formation and pre-existing mediastinal and hilar lymphadenopathy without signs of extraluminal contrast leakage (Fig. 1). Gastroendoscopy was performed and showed a 5-mm tear in the esophagus with an inflow of pus at 33 cm from the incisor teeth. The lymph node culture obtained earlier during EUS-FNA was positive for *Streptococcus parasanguinis*, *Streptococcus salivarius*, and *Bacteroides fragilis*, which were highly sensitive to clindamycin. These bacteria are normally present in the gastrointestinal tract. The pathological report showed granulomatous inflammation.

Since there were no signs of sepsis, we decided to treat him with a combination of antibiotics, esophageal stenting, and minimally invasive video-assisted thoracoscopic surgery (VATS). Five days after re-admission, a 10-cm Ultraflex esophageal stent (Boston Scientific, Marlborough, MA, USA) was placed endoscopically and the posterior mediastinum was drained surgically via right-sided uniportal VATS (u-VATS). The subcarinal lymph node package impeded the view of the tear in the esophagus (Fig. 2). The pleural space was not involved in the abscess, most likely because the subcarinal nodes covered the tear in the esophagus. No further exploration of the subcarinal space was performed to prevent contamination of the thoracic cage, as no abscess formation was observed on CT. Biopsy of the subcarinal lymph nodes was repeated. A 28F tube was placed in the posterior mediastinum. After the esophageal stent was implanted, oral intake was gradually built up.

On postoperative day 9, the chest tube was removed because an upper gastrointestinal X-ray series showed no contrast dye leakage. In the following 7 days, no signs of sepsis were found, and the patient's CRP level (70 mg/L) and WBC count (9.4×10^9/L) decreased. Antibiotics were administered for 28 days. The patient was discharged 15 days after surgery. During further outpatient care (with a total follow-up of 6 months), the patient did not experience any related complications or secondary interventions.

Case 2

Patient B was a 74-year-old woman with a history of breast cancer that had been treated with curative intent 10 years earlier. She was referred with shortness of breath. A CT scan showed multiple lung nodules. In addition, a PET-CT scan showed 4 PET-positive lesions in the right lower lobe, as well as 2 nodules in the right middle lobe and an enlarged PET-positive lymph node in station 7, which were suspicious for metastatic breast cancer lesions. Mammography of the right breast showed no local recurrence of the tumor. EUS-FNA of station 7 was performed using a 19G needle. Directly after the procedure, the patient experienced pain and was admitted for observation. A chest X-ray showed subcutaneous emphysema on the left side of the neck. A non-contrast CT scan showed pneumatosis in

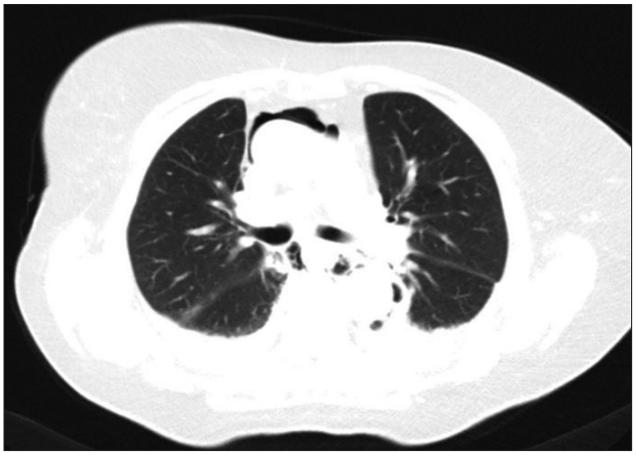

Fig. 3. Computed tomography scan showing pneumomediastinum.

the mediastinum, and pleural effusion and atelectasis on both sides (Fig. 3). The laboratory results demonstrated a CRP of 600 mg/L and a WBC count of 22×10^9/L. The patient was placed on a fasting regimen and intravenous antibiotics were started. The patient experienced clinical deterioration and was admitted to the intensive care unit (ICU) 2 days after the EUS-FNA. She was septic, with a blood pressure of 90/50 mm Hg, a regular heart rate of 110 beats/min, a temperature of 37.7°C, and a respiratory rate of 21 breaths/min. Upper gastrointestinal endoscopy revealed a laceration of the esophagus at 27 cm from the incisor teeth. Consequently an 8-cm colon stent was placed endoscopically. The next day, a chest tube was placed in the left pleural space, draining 400 mL of turbid fluid immediately. Then, u-VATS debridement of a right-sided thoracic empyema was performed and 2 chest tubes were placed in the posterior mediastinum. Postoperatively the patient returned to the ICU. On the following days, the drains kept producing excessive turbid fluid. On day 4 after surgery, upper gastrointestinal endoscopy was repeated; the stent appeared to have been displaced and had to be replaced slightly more proximally. The chest tube in the left pleural space could be removed 6 days after surgery. The next day, another chest CT scan was performed due to an increase in production in the upper chest tube, and demonstrated a small area of fluid collection around the stent and pleural fluid around the lower lobes of both lungs, with atelectasis on the left side. The decision was made to debride the left pleural cavity in combination with opening and draining the posterior mediastinum via u-VATS. Two 28F chest tubes were placed. Because of the size of the esophageal defect and the continuing mediastinal and thoracic leakage, it was decided to laparoscopically place a gastrostomy for gastric decompression and a feeding jejunostomy. Thereafter, a fast recovery and normalization of inflammatory markers was observed. At 14 days after the last surgical procedure, a CT scan with contrast showed no leakage. The gastrostomy could be removed, and the patient was admitted to the rehabilitation center. The esophageal stent was removed 2 months after initial placement. Upper gastrointestinal endoscopy did not show a remaining esophageal defect and the patient returned to a normal diet. The pathology report of station 7 revealed metastatic breast cancer. The patient is currently receiving treatment for metastatic disease.

Discussion

EUS-FNA is an efficient and safe method for assessing mediastinal lymphadenopathies of unknown origin. In the revised guidelines of the European Society for Thoracic Surgeons, EUS or EBUS with FNA is recommended for mediastinal staging [2].

Post-procedural pain and mild hemorrhage are common complications of EUS-FNA. The risk of perforation is low, with a reported incidence of 0.02% [3]. The treatment of esophageal perforation after EUS is mainly based on the treatment principles of Boerhaave syndrome, but it remains unclear whether these are actually similar clinical entities that require similar treatment. Esophageal perforation in patients with Boerhaave syndrome has a poor prognosis and remains a life-threatening condition with a mortality rate of 14.8% [4]. It is a surgical emergency, in which early diagnosis is vital for achieving good outcomes and treatment should be started as early as possible.

The principles of definitive management of esophageal wall rupture are (1) control of the esophageal leak, (2) eradication of mediastinal and pleural contamination, (3) decompression of the esophagus, and (4) nutritional support. The gold standard surgical approach for esophageal perforations is thoracotomy, laparotomy, or both [5]. The traditional open surgical intervention to control the esophageal leak is primary closure with or without autogenous tissue reinforcement, esophageal resection, or exclusion and diversion. To clean the contaminated mediastinal and pleural spaces, tube drainage or simple drainage is performed during the surgical intervention. Anastomotic leakage can be a reason to delay the initiation of oral intake. Nasogastric tube intubation is then performed for enteral nutrition, medication administration, and for gastric decompression. The purpose of surgery is to provide sufficient closure of the defect to allow esophageal healing by following the

aforementioned principles. The location of the injury influences both the surgical approach and the surgical repair options that are chosen. The abdomen needs to be explored if the full extent of the perforation cannot be visualized and extends across the gastroesophageal junction. However, the majority of perforations after EUS are located in the cervical esophagus. Open repair is best performed via a right posterolateral approach. The esophagus is explored, and the defect is repaired by performing a longitudinal myotomy, mucosal repair, closure of the myotomy, and buttress reinforcement with intercostal muscle, pericardial fat, a diaphragm flap, or a pleural patch. Less invasive and endoscopic methods are currently being explored for steps 1 and 2. However, these methods have only been reported after esophageal rupture in patients with Boerhaave syndrome [6], and not for patients with other causes of esophageal perforation. Alternative less invasive treatments consist of a combination of thoracoscopic decortication, drainage, and esophageal stenting. The advantages of the thoracoscopic approach may be shorter thoracic drainage, less postoperative pain, a shorter hospital stay, and a more rapid return to normal function [4,7]. Another method to control the leak is endoscopically assisted mediastinal vacuum therapy. However, nearly all recommendations made for the use of vacuum-assisted closure are based on expert opinions, rather than on scientific evidence.

This paper reports 2 cases of esophageal wall rupture due to EUS-FNA. In both cases, the 4 principles of definitive management of esophageal wall rupture were followed using a minimally invasive approach. In the first case, after eradication of mediastinal and pleural contamination the patient recovered to a considerable extent, and no sepsis occurred. The chest drain was removed on postoperative day 8 and the patient was discharged on postoperative day 15. In contrast, the esophageal leak in the second patient was more difficult to control, and she underwent 2 more operations for additional drainage and nutritional support. The postoperative course was characterized by an oculogyric crisis, excessive production of both drains, and an overall weakened condition.

In the second case, the patient was relatively older and also had distant metastases. These factors are associated with a higher risk of perforation according to the American Society for Gastrointestinal Endoscopy guidelines [8].

In conclusion, to our knowledge, this is the first case report of esophageal wall rupture after EUS-FNA treated with minimally invasive surgery using a combination of esophageal stenting and VATS. Both cases illustrate the need for early recognition and intervention. In conclusion, in selected cases, it is possible to perform minimally invasive surgery for esophageal wall rupture, even in patients with sepsis.

Conflict of interest

No potential conflict of interest relevant to this article was reported.

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