Central nervous system infection and vertebral osteomyelitis as a result of esophageal perforation

A Case Report

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Tammy Ferro DO
1 Banner Good Samaritan Hospital

Abstract

Esophageal injuries in adults are most often iatrogenic, occurring after dilation of esophageal strictures. In this case, a 72 year-old female underwent esophageal dilations for a stricture resulting from radiation for breast and esophageal cancer. She then developed symptoms of pneumonia, followed by seizures and quadraparesis. Imaging revealed esophageal rupture with osteomyelitis, ventriculitis, intraventricular abscess and hydrocephalus from infectious dissemination. Iatrogenic esophageal rupture after dilation has a high mortality; the recommended initial evaluation is with a barium esophogram. Recent experience supports consideration of nonsurgical treatment, which can be as successful as surgical options.

Introduction

Iatrogenic esophageal perforations during endoscopic and surgical procedures account for 57-90% of all the causes of esophageal injuries, occurring at a rate of 0.3% to 4.9% in published series.1-2-3 Esophageal perforations as a result of dilation of malignant strictures are the most common cause of all reported cases of esophageal injuries among adult patients.4 Perforations are far more likely to occur after dilation of malignant strictures, as compared to benign strictures.5 Mortality as a result of esophageal perforation after dilation can be as high as 50%.5

Central nervous system (CNS) infection and vertebral osteomyelitis as a result of iatrogenic esophageal perforation is exceedingly rare: there have been five adult cases of esophageal perforations resulting in CNS complications reported in the literature, with one resulting in osteomyelitis.5-7-8-9 I describe the case of a woman who developed life-threatening complications including a CNS infection and vertebral osteomyelitis as a result of an iatrogenic esophageal perforation after therapeutic dilation of an esophageal stricture.

Case Description

An active 72 year-old woman with an esophageal stricture due to radiation treatment for breast cancer and squamous cell carcinoma of the esophagus underwent periodic dilations for ongoing dysphagia. Weeks after the most recent dilation, she complained of respiratory symptoms and was treated empirically by her primary physician with oral antibiotics. A week later, routine endoscopy demonstrated two mucosal defects proximal to the stricture. A plain-film esophogram revealed barium in the mediastinum, outside the esophagus. Although asymptomatic, she was admitted to the hospital and made nil per os (NPO); empiric antimicrobials were begun. Computerized tomography (CT) scan of the thorax with barium revealed air in the paraspinal tissue and spinal canal, pneumomediastinum, and barium in the mediastinum and airway. Bronchoscopy did not reveal a broncho-esophageal fistula.

After six days she became febrile and comatose. Magnetic Resonance Imaging (MRI) of the brain revealed debris in both lateral ventricles, intraventricular abscess, ventriculitis, and hydrocephalus (Figure 1).
An external ventricular drain was placed for hydrocephalus and administration of intrathecal tobramycin. It was surmised that the esophageal perforation resulted in bacterial contamination of the epidural space and eventually dissemination into the CNS.

On day 13, she developed weakness of the lower extremity paraparesis. Cervical and thoracic MRI was conducted and demonstrated an epidural abscess, osteomyelitis, and compression deformity of multiple vertebrae (Figure 2).

She underwent two hemilaminectomies of which cultures of the epidural abscess were sterile. Her ICU course was challenging. She developed nosocomial infections, profound muscle wasting in both upper and lower extremities, malnutrition, and pulmonary edema/pleural effusions requiring draining chest tube thoracotomies. After two month ICU course, she was transitioned to a prolonged ventilation support facility. She sustained severe cognitive impairment and paraparesis, which is a profound regression from an intelligent woman who played tennis weekly.

**Discussion**

Most cases of CNS involvement reported in the literature are in pediatric patients. In the adult cases, damage of the esophagus complicated by CNS infections is mentioned mostly in patients with malignant strictures undergoing bouginage dilation. Chest pain, confusion, dysphagia, fevers, lethargy, paraesthesias, visual disturbances, nuchal rigidity, tachycardia and tachypnea are all symptoms of infectious dissemination and likely CNS infection. The latter can manifest as meningitis, brain abscesses, vertebral osteomyelitis, and encephalitis as in this patient.

The esophagus, by nature, is a poorly contained organ as it has no protective muscularis layer. The retroesophageal space external to the esophagus consists of prevertebral fascia that is loosely attached to the organ creating septations that prevent the spread of infection, but imparts no protection for the mediastinum. The infection then gains access to the CNS via a valveless conduit of the vertebral venous plexus and azygous vein, both of which drain blood from the vertebrae, esophagus, mediastinum and bronchi.
Early diagnosis is critical to reduce mortality, but about 50% of patients present with symptoms that mimic common diseases such as peptic ulcer, pancreatitis, gastritis and atypical chest pain. The site of perforation can be detected as early as 18 hours with definitive changes on chest X-ray. Subcutaneous emphysema, hydrothorax, pneumothorax, pneumopericardium, pneumoperitoneum, or mediastinal emphysema are found in 90% of patients, but imaging may be negative in the initial hours after perforation.\(^6\)

If significant clinical suspicion for esophageal perforation exists, a contrasted esophagram should be performed. Gastrograffin is a good initial contrast agent, as it does not form granulations if leaked into the mediastinum as barium would. If aspirated, however, gastrograffin can cause necrotizing pneumonitis. This is not true of barium and the dense nature of barium imparts a superior ability to permeate smaller perforations. Contrasted CT can further characterize leaks and localize abscesses and fistulas. Pneumomediastinum is the most common finding seen with CT scanning. Endoscopic visualization offers both diagnostic and therapeutic advantages, but should not be used in the acute setting as air insufflation during the procedure may dissect within the wall of the esophagus thereby worsening subcutaneous emphysema.

Oropharyngeal bacteria are most often implicated in infectious complications after esophageal perforation. Contamination of the dilator during its descent into the esophagus by oropharyngeal organisms such as \textit{S. aureus} and \textit{Streptococcus} species are most commonly encountered in the CSF, intracranial and mediastinal abscesses, and blood.

Until the 1980's, esophageal perforation was a surgical emergency and repair or resection was indicated within 24 hours to prevent morbidity and mortality. In the last two decades, however, esophageal perforations have been managed successfully with endoscopic placement of esophageal stents. Survival rates have been reported to be 60-90% for non-operative treatment, which rivals that of surgically managed patients.\(^7\) Our patient suffered such a catastrophic sequela due to her esophageal perforation that surgical repair could not be contemplated.

Antimicrobial agents of choice should encompass anaerobes, gram-negative and gram-positive aerobes. Antimicrobial prophylaxis is controversial. Expert consensus does not advise the routine use of peri-procedure antibiotics for esophageal dilations. One author recommended prophylactic intravenous broad-spectrum antibiotics immediately before and after the dilation up to 24 hours.\(^8\) Their justification for this short regimen is that the bacteremia is only transient.

Regardless of the mode of treatment, careful observation, withholding of oral feeding while initiating parenteral alimentation, oral or naso-esophageal suction, antimicrobial administration, and gastric acid reducing medications remains standard of initial care. Broad-spectrum antimicrobials (covering gram negative, gram positive, and anaerobic bacteria) are advocated as soon as esophageal injury is suspected.

References