by the ipsilateral inferior pulmonary vein. This shunt may exacerbate symptoms of heart failure in patients with existing intracardiac anomalies. These patients, as our case illustrates, usually have continuous murmurs located over the chest, back, or axilla on the affected side. Due to the high prevalence of associated cardiac anomalies, it is unclear whether the symptoms of heart failure are due to the volume overload from the sequestration or the underlying heart disease.

To our knowledge, there have been only two previous cases of CHF associated with ILS and normal intracardiac anatomy. White and associates reported a 10-week-old infant whose symptoms of CHF improved after lobectomy; however, the patient died of acute tracheobronchitis five weeks postoperatively. Ransom and associates reported a 6-month-old infant who initially presented at 4 months of age with CHF. The diagnosis of ILS was demonstrated with an aortogram. Our patient who had CHF associated with ILS is the first case to be diagnosed in the neonatal period. Presumably, the patient's prematurity contributed to the early presentation of CHF. The diagnosis of pulmonary sequestration in this case was suggested by two-dimensional echocardiography in conjunction with Doppler flow analysis. Recently, Doppler color flow mapping has been reported to be useful in the evaluation of such vascular lesions.

In conclusion, we describe the case of a neonate who presented with CHF from ILS but who did not have an associated cardiac anomaly. We found two-dimensional echocardiography and pulsed-Doppler analysis helpful in suggesting the diagnosis of pulmonary sequestration. This finding of a pulmonary sequestration was confirmed by aortography, which is currently the best diagnostic tool for this disorder. Surgical lobectomy or segmentectomy has been curative in nearly all cases with hemodynamic symptoms, but the decision to resect the lesion requires careful consideration of the volume load on the heart, the presence of underlying cardiac defects and the vascular supply to the sequestrum, since there is some mortality associated with lobectomy.

REFERENCES


Diagnostic Value of Pleural Fluid Cytology in Occult Boerhaave's Syndrome

Michael Drury, M.D.; William Anderson, M.D.; and John E. Heffner, M.D., F.C.C.P.

When Boerhaave's syndrome presents with atypical clinical features and eludes prompt diagnosis, delays in surgical therapy increase complications and mortality. We present a patient with occult Boerhaave's syndrome who had nondiagnostic esophageal contrast studies and thoracic computed tomography. Pleural fluid cytologic analysis established the presence of esophageal rupture by detecting undigested food particles. (Chest 1992; 102:976-78)

Despite more than 250 years of clinical experience with the diagnosis, Boerhaave's syndrome (spontaneous esophageal rupture) remains a potentially lethal and frequently elusive medical condition. Prognosis rapidly diminishes when diagnosis and surgical repairs are delayed with mortality rates reported as high as 100 percent in some series after one week of nonsurgical medical care. Unfortunately, patients frequently present with a "nonclassical" history without an episode of preceding emesis, and results of the physical examination and laboratory studies may provide atypical, nondiagnostic clues. We report the case of a patient with spontaneous esophageal rupture that remained undiagnosed despite a thorough evaluation with contrast esophagography, thoracic computed tomography (CT), and routine microscopic examination of pleural fluid for food particles. The diagnosis was confirmed by detection of undigested food particles in the pleural fluid cytologic preparation.

CASE REPORT

A 64-year-old man presented 3 h after the sudden onset of left posterior chest pain that started while lifting a heavy object and radiated to his left shoulder and abdomen. The patient had no history of productive cough, fever, chills, or dyspnea before the onset of pain. He denied alcohol abuse, dysphagia, vomiting, or loss of consciousness. Physical examination revealed a thin man with mild respiratory distress and restlessness from thoracic pain. Blood pressure was 140/70 mm Hg, pulse rate was 88 beats per minute, respiratory rate was 30/min, and temperature was 36.7°C. Subcutaneous crepitation was absent. Chest auscultation revealed dimin-

*From the Departments of Medicine (Drs. Drury and Heffner) and Pathology (Dr. Anderson), St. Joseph's Hospital and Medical Center, Phoenix.

Reprint requests: Dr. Heffner, St. Joseph's Hospital, 350 West Thomas, Phoenix 85013
ished breath sounds, egophony, decreased tactile fremitus, and dullness to percussion at the left lung base. The abdominal examination revealed mild left upper quadrant tenderness but no guarding or rebound. Laboratory studies showed a white blood cell count of 17,000 cells per microliter, with 84 percent granulocytes and 9 percent band forms. Serum amylase level was normal. Arterial blood gas analysis on 2 L O2/min flow by nasal cannula showed a pH of 7.39, PaO2 of 67 mm Hg, and a PaCO2 of 31 mm Hg. The hospital admission chest roentgenogram demonstrated severe kyphoscoliosis with a left lower lobe alveolar infiltrate and left-sided pleural effusion (Fig 1). A small left pneumothorax was also present, but no signs of pneumomediastinum or subcutaneous emphysema existed.

Several hours after hospital admission, decubitus chest roentgenograms showed a large free-flowing left pleural effusion with marked increase in size of the left pneumothorax. A left chest tube was urgently placed that drained 1 L of dark, turbid fluid that had a white blood cell count of 389 cells per microliter, protein of 3.2 g/dl (serum protein, 6.0 g/dl), glucose of 319 mg/dl, lactate dehydrogenase of 5,060 IU/L, amylase of 496 IU/L, and pH of 6.65. Gram stain and wet-preparation of pleural fluid examined by microscopy for vegetable matter were negative. Pleural fluid was sent for culture and cytology.

The initial differential diagnosis included both necrotizing bacterial pneumonia with pyopneumothorax and spontaneous esophageal rupture. The latter diagnosis was considered despite the patient's denial of emesis because of the increased pleural fluid amylase content. Subsequently, a family member recalled that the patient experienced a severe episode of vomiting 24 h before the onset of chest pain. A thoracic CT scan revealed a large left posterior pleural fluid collection and parenchymal consolidation of the left lower lobe. There was no evidence of mediastinal air or communication of pleural fluid with the esophagus. Contrast esophagography performed with the patient in the left lateral decubitus position using first meglumine diatrizoate (Gastrografin) and then barium sulfate failed to demonstrate extravasation of contrast into the mediastinum. A thoracentesis directed at a nondrained loculum of pleural fluid revealed a pH of 7.03 and amylase level of 220 IU/L.

Despite broad-spectrum antibiotics and placement of a second left chest tube, the patient's condition deteriorated with manifestations of sepsis syndrome and he required intubation, mechanical ventilation, and vasopressor therapy. Blood and pleural fluid cultures grew Pseudomonas aeruginosa, which prompted adjustment of antibiotics to include gentamicin and mezlocillin. On the third hospital day, the pleural fluid cytologic studies showed the presence of undigested meat and vegetable matter (Fig 2 and 3). A left thoracotomy performed the same day revealed a 5-mm tear in the distal intrathoracic esophagus, which was repaired with a gastric fundal patch.

A prolonged hospital course ensued complicated by Pseudomonas pneumonia, respiratory failure, and Burkholderia glabrata infection of the pleural space. The patient's condition improved, however, and he was discharged home after a nine-week hospitalization. He had returned to his normal state of health when examined five months later.

**Discussion**

Spontaneous esophageal rupture is one of the most rapidly fatal conditions associated with perforation of the gastrointestinal tract. Nearly 45 years after the first reported success with surgical repair, thoracotomy with primary closure of the esophageal tear remains the most effective therapeutic approach. Mortality and morbidity, however, appear in most studies to correlate directly with the length of delay from onset of symptoms to diagnosis and definitive care.¹

Eliciting a history from the patient or family of forceful

![Figure 1](http://journal.publications.chestnet.org/pdfaccess.ashx?url=/data/journals/chest/21654/)

**Figure 1.** Hospital admission chest roentgenogram showing severe kyphoscoliosis, left lower lobe infiltrate, left pleural effusion, and left pneumothorax (arrow).

![Figure 2](http://journal.publications.chestnet.org/pdfaccess.ashx?url=/data/journals/chest/21654/)

**Figure 2.** Pleural fluid cytologic specimen prepared by cytocentrifugation showing a particle of undigested meat and cross striations of skeletal muscle (arrow) (Papanicolaou stain, × 400).

![Figure 3](http://journal.publications.chestnet.org/pdfaccess.ashx?url=/data/journals/chest/21654/)

**Figure 3.** Cytologic preparation of pleural fluid showing vegetable matter with chitin cell walls (arrows). These particles were apparent in numerous regions of each microscopic field (Papanicolaou stain, × 100). Inset shows a vegetable cell wall examined by polarized light (arrow) (Papanicolaou stain, × 400).
vomiting preceding the onset of thoracic symptoms is central to a rapid diagnosis. Unfortunately, the initial clinical manifestations of Boerhaave's syndrome may commonly present in atypical ways obscuring the diagnosis and delaying surgical care. The classic presentation with a history of severe vomiting after a binge of eating and alcohol ingestion, lower thoracic pain, subcutaneous emphysema, and respiratory distress rapidly progressing to prostration may appear in less than 50 percent of instances. In the remaining patients, initial manifestations may simulate other conditions, such as peptic ulcer disease, myocardial infarction, or pancreatitis.

The sudden onset of thoracic pain with roentgenographic signs of a lower lobe infiltrate, pleural effusion, and small pneumothorax, in our patient who initially denied emesis, simulated a necrotizing pneumonia with rupture of an intraparenchymal abscess into the pleural space. The pleural fluid analysis, however, suggested the possibility of Boerhaave's syndrome based on the moderately elevated amylase value and low pH—characteristic but nonspecific features of esophageal rupture.

Despite early clinical suspicion and urgent diagnostic pursuit of esophageal rupture, a carefully planned laboratory evaluation may nevertheless fail to confirm the diagnosis as occurred in the present report. Although Boerhaave's syndrome frequently demonstrates left-sided pleural effusions associated with pneumothorax, auscultatory and roentgenographic evidence of mediastinal emphysema are more suggestive findings. Mediastinal emphysema was absent in the present patient as reported in 10 to 12 percent of patients. Contrast studies of the esophagus performed with the patient in the left lateral decubitus position, initially with meglumine diatrizoate (Gastrografin) followed if negative by barium sulfate, are indicated in all instances of suspected rupture. Although considered relatively sensitive, previous reports of false-negative results of examinations as occurred in this patient exist with instances of normal meglumine diatrizoate studies reported as high as 10 to 50 percent of patients with esophageal rupture. Detection of the esophageal perforation may be enhanced by repeating the contrast study after several hours.

Thoracic CT offers potential value in Boerhaave's syndrome by demonstrating free mediastinal air and open communication of the esophagus with mediastinal structures. Limited experience, however, does not yet define the sensitivity and diagnostic accuracy of this test, which was negative in the patient whose case was reported. Drainage through a chest tube of previously swallowed "tracer" fluids, such as antacid, charcoal, or methylene blue, may confirm the diagnosis in some patients. Fractionation of pleural fluid amylase with detection of the salivary component may confirm esophageal rupture, but the delay in results of analysis for many clinical laboratories diminishes this test's utility. The safety and sensitivity of esophagoscopy remain unestablished in the presence of an esophageal tear.

The present patient remained clinically perplexing until results of pleural fluid cytologic evaluation demonstrated the presence of undigested meat and vegetable matter. Although previous reports indicate that food particles in the pleural space are diagnostic of esophageal rupture, their presence frequently goes undetected in the Gram stain and wet preparations recommended for analysis. The pleural fluid smear prepared by routine centrifugation in the present report failed to demonstrate meat or vegetable matter despite careful scrutiny directed by the high clinical suspicion for Boerhaave's syndrome. In contrast, the plethora of food particles that were easily detected on the cytologic slides indicate that high-speed cytocentrifugation concentrates the undigested debris thereby enhancing the sensitivity of the technique. The ability of most clinical laboratories to complete pleural fluid cytologic analysis within several hours of specimen submission warrants consideration of this diagnostic technique in patients with clinical suspicion of esophageal rupture.

ACKNOWLEDGMENTS: The authors thank Cynthia Bockow for excellent secretarial assistance and Kay Wellik for library services.

REFERENCES