CASE REPORT

Broncho-pleuropericardial fistula complicating staphylococcal sepsis

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ABSTRACT

This is a rare case of broncho-pleuropericardial fistula in a 12-year-old female who presented with fever, painful joint swelling, and pleural and pericardial effusion secondary to disseminated methicillin-sensitive *Staphylococcus aureus* infection. The pleural and pericardial effusion were drained, however, air leak was observed from both tubes and was synchronous with mechanical inspiration. A broncho-pleuropericardial fistula was suspected and confirmed with computed tomography. This case report demonstrated that disseminated *S. aureus* bacteremia could result in broncho-pleuropericardial fistula. The ability of disseminated staphylococcal infection to produce pnemopericardium should be added to the list of other complications associated with disseminated staphylococcal sepsis.

Key words: Bronchial fistula, fistula, pericardial tamponade, staphylococcal infections

INTRODUCTION

Bronchopleural fistula is a condition that occurs with low frequency and is usually seen after trauma or thoracic surgery. In contrast, pericardial fistula is a rare condition that has been described in association with various disease states.[1-4] When the fistulous communication occurs between the pericardium and either the bronchus, [2] bowel, [4] or esophagus, [5] it results in pneumopericardium that could potentially lead to cardiac air tamponade and hemodynamic instability.^[6] All previous reports of pericardial fistulae have described a communication between the pericardial sac and a single other viscus. This is a report of a very rare fistulous communications between the pericardium, the right bronchus, and the right pleural cavity in a child with septic shock secondary to methicillin-sensitive staphylococcal bacteremia. Written consent for publication was obtained from the parents.

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CASE REPORT

A 12-year-old female, previously healthy, was admitted from the emergency department to the intensive care unit with a two weeks history of fever and multiple asymmetrical joint swelling and pain. Her admission temperature was 39.6°C, heart rate (HR) 130 beats/min, arterial blood pressure (BP) 127/57 mmHg, respiratory rate (RR) 35 breaths/min, and oxygen saturation (SpO₂) 93% on air. On examination, the patient had hair lice with multiple scratch marks on her scalp, bilateral pleural effusion, ejection systolic murmur grade 2/6 over the aortic area, tender hepatomegaly (3 cm below the costal margin, liver span 10 cm), and multiple joint swelling, redness, and tenderness involving the right middle finger, right hip, and both knees and ankles. Admission laboratory results revealed a white blood cell (WBC) count of 6000 cells/mm³, lymphocytes 0.9×10⁹ cells/l, granulocytes 4.5×109 cells/l, monocytes 0.2×109 cells/l, reticulocytes 28×10° cells/l, platelets 156,000 cells/mm³, hemoglobin 107 g/l, serum sodium 125 mmol/l, serum potassium 4.8 mmol/l, serum glucose 4.3 mmol/l, serum urea 20.7 mmol/l, serum creatinine 90 µmol/l, lactate dehydrogenase (LDH) 1427 IU/L, aspartate aminotransferase (AST) 408 IU/L, alanine transaminase (ALT) 307 IU/L, alkaline phosphatase (ALP) 184 IU/L, albumin 19 g/l, gamma-glutamate transferase (GGT) 110 IU/L, creatine kinase (CK) 142 IU/L, total bilirubin 145 μmol/l, serum osmolality 289 mOsm/kg, and urine osmolality 647 mOsm/kg. The patient's international normalized ratio (INR) was 1.2, prothrombin time (PT) 14 (control 12), and activated partial thromboplastin time (aPTT) 36 (control 34). Screening tests for sickle cell disease, hepatitis A, hepatitis C, and human immunodeficiency virus were negative; however, she tested positive for anti-hepatitis B surface antigen (anti-HBs Ag) antibodies. Chest X-ray confirmed the presence of bilateral pleural effusion (right > left) and showed widened mediastinum with a pear-shaped heart suggestive of pericardial effusion. A twelve-lead electrocardiogram (ECG) demonstrated sinus tachycardia, diffuse ST-segment elevation with generalized T-wave inversion. Transthoracic echocardiogram revealed moderate size pericardial effusion and no evidence of endocarditis or valvular heart disease. Blood, sputum, and urine cultures were obtained in the emergency department and the patient was started empirically by the emergency department physician on ceftazidime 1.3 g intravenous (IV) every 8 hours and flucloxacillin 1.2 g IV every 6 hours. In addition, a right chest tube thoracostomy was inserted and placed to underwater seal; 800 ml of serous fluid was drained and there was no air leak. Analysis of pleural fluid showed lactate dehydrogenase (LDH) 5 IU/L, albumin 19 g/L, and pH 7.37. Gram-positive cocci in clusters were seen on Gram staining of the pleural fluid (admission sample). The patient's hair was shaved and she was started on anti-lice shampoo. Sputum microscopy was negative for acid-fast bacilli and a sample for mycobacterium tuberculosis culture and polymerase chain reaction test was sent off.

On the second day, the patient became more tachypneic (RR 40 breaths/min) and hypotensive (BP 90/40 mmHg). Her trachea was intubated, lungs were mechanically ventilated, antibiotics were changed to meropenem to broaden her coverage, and she was started on dopamine and norepinephrine infusions. After initiation of mechanical ventilation, air leak from the right chest tube was observed suggesting the presence of bronchopleural fistula, however, she maintained her SpO₂ > 95% on FiO₂ 0.4. Echocardiography-guided pericardiocentesis was performed and 500 ml of pus was aspirated and a pig-tail catheter was inserted into the pericardium. Pericardial fluid analysis showed proteins 42 g/l, albumin 27 g/l, and Gram positive cocci in clusters on microscopy. Chest X-ray showed right minimal hydropneumothorax. Ultrasound examination of the abdomen revealed perinephric and subhepatic collections with free fluid in the abdomen and pelvis. Methicillin-sensitive Staphylococcus aureus was cultured from sputum, blood, pleural fluid, and pericardial aspirate. Urine cultures were negative. Rheumatoid factor levels were

slightly above the upper range of normal at 48.8 IU/L. Cytomegalovirus IgM antibodies were negative, but the IgG titer was 1/21,000 IU/L.

On day 4, air started to leak continuously through the pericardial pig-tail catheter which was still draining pus and the air leak was most marked during mechanical inspiration. A broncho-pericardial or broncho-pleuropericardial fistula was suspected. The pig-tail catheter was inadvertently clamped and the patient's BP decreased to 80/50 mmHg and HR increased to 120 beats/min. BP and HR returned to baseline values after de-clamping the catheter. Computed tomography (CT) of the chest showed pneumopericardium [Figures 1 and 2] and hydropneumothorax [Figure 2]. The radiologist injected radio-opaque contrast material easily into the pericardial pig-tail catheter and the contrast material immediately spilled over into the right pleural cavity [Figure 3]. A CT fistulogram, thus, confirmed the

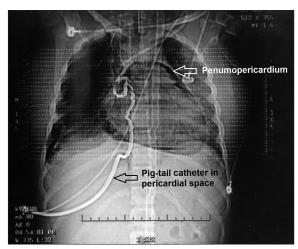


Figure 1: Scout image of the chest showing right pneumothorax, pneumopericardium, and a pig-tail catheter overlying the right cardiac border

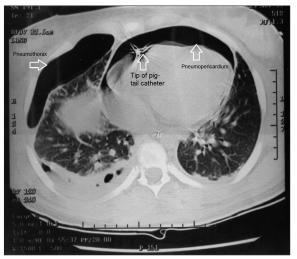


Figure 2: Axial image of the lower chest (lung window) showing moderate size right hydropneumothorax and pneumopericardium with the tip of the pig-tail catheter located in the anterior pericardial cavity

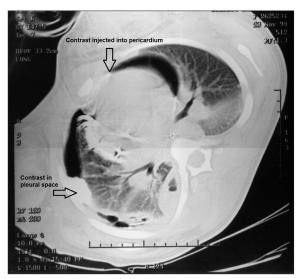


Figure 3: Image obtained post-contrast injection through the pig-tail catheter that demonstrated accumulation of the contrast within the pericardial and right pleural cavity

presence of communication between the right pleural space and the pericardium [Figure 3]. The cardiothoracic surgeon recommended conservative management given the patient status at the time. On day 5, the abdominal collections were aspirated laparoscopically. On day 11, the patient developed disseminated intravascular coagulation and was bleeding from everywhere. On day 13, she died of multi-organ failure and her parents refused an autopsy.

DISCUSSION

All previous reports of pericardial fistulae have described a single communication between a viscus and the pericardial sac. In this report, the patient had fistulous communications between the pericardium, a bronchus, and the right pleural cavity as a complication of staphylococcal bacteremia. Although the actual fistulous tracts were not visualized on radiological examination, the spillage of the contrast material into the right pleural space [Figure 3] immediately after injection of the contrast into the pericardial space through the pig-tail catheter provided sufficient evidence for the existence of a pleuro-pericardial fistula. Since the contrast material is extremely irritant to the lung, it was not possible to inject contrast into the tracheobronchial tree to demonstrate the fistulous communication between the bronchial tree and the pericardium. However, the persistent air leak through the pericardial catheter that was synchronous with mechanical inspiration despite the presence of a right thoracostomy tube strongly supported the presence of such communication. Furthermore, a rim of pneuomopericardium was persistently observed [Figures 1-3]. It is extremely unlikely that air was escaping from the right pleural space into the pericardium as air travels down the path of least resistance which in this case was the patient's right chest tube. Instead, air was likely pushed down a communication tract between the tracheobronchial tree and the pericardium. In fact, the decrease in arterial blood pressure in response to the inadvertent clamping of the pericardial catheter and the return of blood pressure to baseline value immediately on unclamping of the catheter suggested the development of transient cardiac tamponade with air being trapped in the pericardial sac during mechanical ventilation. Unlike pneumothorax, isolated pneumopericardium is not treated with tube drainage unless it is accompanied by pericardial effusion and/or there are signs of pericardial tamponade. [7] In the current patient, tension pneumopericardium developed momentarily when the pig-tail catheter was inadvertently clamped and it resolved after unclamping of the tube.

Necrotizing respiratory infections may occasionally result in a bronchopleural fistula and rarely in a bronchopericardial fistula. Such infections include pulmonary tuberculosis,[8] necrotizing pneumonia, and invasive pulmonary aspergillosis.[7] In this report, however, the patient had methicillin-sensitive S. aureus bacteremia leading to septic shock and eventually multi-organ failure. The bacteremia was most likely due to infected scalp wounds secondary to heavy infestation with hair lice and the consequent itching and scratching associated with it. The patient was immunocompetent and had no chronic underlying diseases prior to her presenting illness. The initial choice of antibiotics were made by the emergency department physician and was not changed on arrival to the intensive care unit given the results of the Gram stain and the fact that community-acquired methicillin-resistant S. aureus is uncommon in our setting. However, when the patient's condition worsened on the following day, the antibiotics were changed to meropenem due to its broader spectrum activity. This decision was made in consultation with an infectious disease specialist. In addition, culture results showed that the staphylococci isolated from all sites were sensitive to meropenem.

It is unlikely that the fistulous communication was iatrogenic following the pig-tail catheter insertion since the procedure was done under echocardiographic guidance and the air leak occurred two days later. Although it followed the initiation of mechanical ventilation, it is unlikely that this complication was secondary to ventilator-induced lung injury since the patient was ventilated with pressure-control ventilation as customary in children. The pericarditis that the patient had (based on persistent generalized ST-elevation in the ECG) was also most probably caused by her *S. aureus* bacteremia as this was the only organism isolated from all cultures including pericardial fluid. In keeping with this is the fact that in multivariable analysis,

infective endocarditis caused by *S. aureus* has been identified as an independent predictor of mortality.^[9] In conclusion, broncho-pleuropericardial fistula should be considered in the differential diagnosis of patients who develop pneuomothorax and pneumopericardium, with/without sudden unexplained hypotension.

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