Spontaneous Pneumothorax in Children with AIDS*

Scott A. Schroeder, MD; Debra Beneck, MD; and Allen J. Dozor, MD

The incidence of pneumothorax in HIV-infected children has not been reported. In adults with AIDS, pneumothorax has been described exclusively in association with *Pneumocystis carinii* pneumonia (PCP). We report the cases of three children with AIDS, one with lymphoid interstitial pneumonitis (LIP) without evidence of PCP and two with PCP, all of whom developed spontaneous pneumothorax (SP). On presentation, none of the children had any risk factors for the development of pneumothorax, but all had radiographic evidence of subpleural cystic lesions and bilateral pleural adhesions. None of the patients responded to conservative medical management, which included chest tube thoracotomy and chemical pleurodesis. Two patients underwent pleurectomy that resulted in resolution of the pneumothorax. Both patients with PCP who developed pneumothorax died, but the patient with LIP and SP has had no recurrences of any serious respiratory problems 3 years after pleurectomy and excision of the intrathoracic cysts.

(CHEST 1995; 108:1173-76)

LIP=lymphoid interstitial pneumonitis; PCP=*Pneumocystis carinii* pneumonia; SP=spontaneous pneumothorax

Key words: pediatric AIDS; pneumothorax; subpleural cysts

Pulmonary disease is a major contributor to the morbidity and mortality of pediatric HIV disease. The incidence of *Pneumocystis carinii* pneumonia (PCP), lymphoid interstitial pneumonitis (LIP), and bacterial pneumonias in HIV-infected children has been well documented, but there are no reports of spontaneous pneumothorax (SP) associated with pediatric HIV infection.1-4 In adults with AIDS, the development of pneumothorax, bronchopleural fistulae, and other air leaks has been most commonly associated with PCP, and the incidence of pneumothorax secondary to PCP has been reported to be as high as 9%.5-7 Adults with AIDS who develop air leaks have a grim prognosis, with poor short-term survival. There is no consensus as to the most effective treatment.7-11 We report the cases of three children with HIV infection, one with LIP and two with PCP, who developed SP refractory to standard medical therapy.

REPORT OF CASES

**Case 1**

A 14-year-old girl was admitted to the hospital with a 3-day history of dry cough, and a 1-day history of midsternal chest pain but without any evidence of respiratory distress. Two years prior to admission, HIV infection was diagnosed. At that time, she had a chest roentgenogram which showed bilateral interstitial disease without evidence of cysts, bullae, or pneumatoceles. In the year prior to her hospitalization, she had had no lung disease but she had recurrent herpes zoster dermatitis and was receiving acyclovir prophylaxis. She was on an experimental protocol with dideoxyinosine; this medication was discontinued 3 months prior to admission to the hospital because of pancreatitis. She also took fluconazole for oral candidiasis and prophylactic trimethoprim-sulfamethoxazole.

A chest roentgenogram obtained on the day of admission demonstrated pneumothorax of 30% and 15% on the left and right sides, respectively. Bilateral chest tubes attached to suction drainage were inserted and the lungs were fully reexpanded. *P carinii* was demonstrated on methenamine-silver stained and was cultured from her sputum; therapy was begun with intravenously administered trimethoprim-sulfamethoxazole and cefuroxime, orally administered prednisone, and 50% oxygen by face mask. Over the next 10 days, the pneumothorax reappeared despite the placement of numerous chest tubes. On hospital day 38, chemical pleurodesis with doxycycline was attempted but was unsuccessful. A CT scan of her thorax demonstrated diffuse airspace disease with cystic changes in the upper lung fields bilaterally. On hospital day 49, she underwent left thoracotomy, partial pleurectomy, and mechanical pleurodesis. Postoperatively, both lungs reexpanded, and she was extubated; instances of pneumothorax did not recur. On hospital day 51, fiberoptic bronchoscopy was done because of persistent left lower lobe atelectasis. Thick secretions were found, but no microorganisms were seen or cultured from that fluid. On hospital day 53, she had been weaned to an FiO2 of 28%, and there was no recurrence of the pneumothorax as shown on the roentgenogram of the chest. Later that day, she developed generalized tonic-clonic seizures that were unresponsive to anticonvulsants, metabolic acidosis, and hypoxemia. No postmortem consent was obtained. Her death certificate listed cardiopulmonary arrest as a consequence of seizures and AIDS as the cause of death.

Histologic findings of the biopsy specimen of lung tissue demonstrated a mild interstitial inflammatory process with patchy fibrosis and calcification, mild emphysema, and recent intra-alveolar hemorrhage. Granulation tissue with foreign body reaction extended into the pleura (Fig 1). Stains as well as cultures of lung tissue, lymph nodes, and pleural fluid for bacteria, fungi, mycobacteria, viruses, and for *Pneumocystis* organisms were negative. There was no histologic evidence of viral infection such as cytomegalovirus, measles, Epstein-Barr virus, or herpes simplex virus.

**Case 2**

The patient was a 9-year-old girl admitted to the hospital with a 2-day history of cough and right pleuritic chest pain. A diagnosis of AIDS and PCP was made on the basis of a lung biopsy 7 years prior to admission to the hospital. She had a history of pneumococcal sepsis, cardiomyopathy, partial complex seizure disorder, and hyperreactive airway disease, but she had no history of opportunistic infections. Three months prior to admission, she had a right-sided pneumothorax that resolved after chest tube drainage. Two weeks prior to admission, she had another right-sided pneumothorax. She underwent chest tube thoracotomy and quinacrine pleurodesis and was discharged after a 9-day hospitalization. At the time of discharge, roentgenogram of the chest showed no pneumothorax but numerous cysts in the right middle and right lower lobes. At the time of this admission, she was tachypneic and had nasal flaring, intercostal retractions, bilateral coarse crackles, and decreased breath sounds over the right hemithorax. She was taking zidovudine, digoxin, furosemide, trimethoprim-sulfamethoxazole, and theophylline. A chest roentgenogram revealed a 40% right-sided pneumothorax. Staining and culture of induced sputum and
pleural fluid produced no evidence of bacteria, fungi, viruses, or *P. carinii*. She was given 100% oxygen via a nonrebreathing mask and cefuroxime intravenously, and a chest tube was placed. By hospital day 4, she needed only 2 L/min of oxygen via nasal cannula to keep her oxygen saturation greater than 96%, but the pneumothorax persisted. On hospital day 7, she underwent tetracycline pleurodesis of the right pleural space; the lung did not fully expand. A CT scan of the thorax showed bullae in all of the lobes of the right lung (Fig 2). On hospital day 13, a right thoracotomy was performed. Numerous cysts were excised from the right middle and lower lobes. Cultures and stains of the lung tissue for bacteria, fungi, protozoa, and viruses were negative. Histologic examination of the lung tissue showed a dense lymphocytic infiltrate of the interstitium with peribronchial lymphoid nodules and germinal centers, as seen in LIP. The pleura was focally fibrotic with granulation tissue, foreign body reaction, and calcification. There were no viral inclusions or cellular changes suggestive of acute viral disease. She was extubated on hospital day 16 and discharged on hospital day 21. Since discharge from the hospital, she has had no recurrences of pneumothorax, nor has she had any other major respiratory difficulties.

**Case 3**

The patient was a 10-year-old boy admitted to the hospital with a 2-day history of nonproductive cough and fever and a 1-day history of sternal chest pain and tachypnea. A diagnosis of AIDS was made six years prior to admission to the hospital. He had a history of chronic diarrhea of unknown cause, failure to thrive, intermittent epistaxis, and splenomegaly; he had no history of lung disease. Eight months prior to admission during an evaluation for a fever, *Mycobacterium avium* was cultured from his blood. After it had been cleared from his blood, a Broviac catheter was placed. Three weeks prior to admission, he had an episode of hematemesis; a roentgenogram of the chest at that time showed no evidence of lung disease. His medications included granulocyte colony-stimulating factor, erythropoietin, fluconazole, ciprofloxacin, and clarithromycin. He was allergic to trimethoprim-sulfamethoxazole and penicillin.

On admission, he was in moderate respiratory distress with a respiratory rate of 50 breaths per minute, perioral cyanosis, intercostal retractions, and bibasilar crackles. A roentgenogram of the chest revealed bilateral interstitial infiltrates. The oxygen saturation was 82%. Therapy was initiated with oxygen, 10 L/min via face mask as well as intravenously administered pentamidine, cefotaxime, acyclovir, and ciprofloxacin. Bronchoalveolar lavage was deferred because of his poor general condition. His clinical condition worsened; a chest roentgenogram performed 36 h after admission to the hospital revealed bilateral pneumothoraces. He was intubated, placed on mechanical ventilation, and underwent surgical drainage with bilateral chest tubes. PCP was confirmed by methenamine-silver staining and culture of the organism in bronchial lavage specimens; no other organisms were seen on either stain or culture of the respiratory secretions. By hospital day 9, the pneumothorax on the left had resolved, and he had been weaned to an airway support system (BiPAP; Respironics, Inc; Murrysville, Pa) with an oxygen flow of 5 L/min; intermittent positive airway pressure of 8; expiratory positive airway pressure (EPAP) of 4; and a respiratory rate of 12 breaths per minute. A chest roentgenogram showed bilateral interstitial and alveolar infiltrates and persistence of the right-sided pneumothorax. On hospital day 12, he was transferred to the general pediatric wards. On hospital day 20, he became tachypneic, lethargic, and had oxygen desaturation to 85% despite respiratory support with BiPAP and supplemental oxygen. A chest roentgenogram revealed that the right-sided pneumothorax persisted, and he had developed pneumomediastinum. Since his family had requested that no extraordinary support measures be initiated, comfort measures were given and he died on hospital day 24. No postmortem consent was obtained.

**Discussion**

Pulmonary disease is a major contributor to the morbidity and mortality of children with AIDS, but to date, the incidence of pneumothorax in the pediatric AIDS population is unknown.1-5 In children with congenital immunodeficiencies, SP is common and is associated with PCP. This association was first described more than 30 years ago; the mor-

**Figure 1.** Lung biopsy of patient in Case 1. The pleura (P) is separated from the underlying pulmonary tissue, which shows scarring, dystrophic calcification, chronic inflammation, and a foreign body reaction (arrow). Air and blood occupy the emphysematous space between the pleura and the parenchyma (asterisk) (hematoxylin and eosin, original magnification x40)

**Figure 2.** Patient in Case 2. CT scan shows bilateral cystic changes. The cysts were more pronounced on the right but were present in all lobes.
tality then was 100%. Today, the prognosis of pediatric patients with PCP and SP in this setting is good when the patients are treated with parenterally administered pentamidine and, if necessary, thoracotomy.14

The pathogenesis of pneumothorax secondary to PCP in adults is unknown but is thought to be associated with the rupture of subpleural cavities or emphysematous blebs.17-21 Radiographic evidence of bullae and cysts, though common in adults with PCP, is an unusual finding in children with PCP. In recent reports on the pathologic findings in the lungs of children with AIDS, no mention is made of subpleural cysts associated with PCP.2,4,22,23

In adults with AIDS, the frequency of SP has been increasing and has been most often associated with PCP.5,6 When adults with AIDS develop pneumothorax, it tends to recur. There is no consensus about the most effective treatment, and the long-term outlook is grim.8,11 Numerous modalities have been used to treat SP in adults with AIDS. Some authors have reported success with chemical pleurodesis.11 For the majority of patients, conventional therapy with intercostal tube drainage with or without pleurodesis fails to eradicate the air leak.2,3,5,6 One report on the use of parietal pleurectomy for the treatment ofbronchopleural fistulae in patients with AIDS showed resolution of the air leaks with no recurrence of ipsilateral pneumothorax. Of the 14 patients who underwent pleurectomy in that study, 6 were reported as still living.11

The causes of the pulmonary pathologic findings of the three patients in our report differed, but two had chronic radiographic changes, parenchymal cysts contiguous with the pleura, and extensive pleural adhesions. Histologically, both had interstitial inflammation extending to the pleura, but in neither was there evidence of active infection at the time of pleurectomy. Interstitial inflammation in the lungs of HIV-infected children may lead to the formation of cysts and air leaks that may predispose the damaged lung to pneumothorax. The chronic inflammation would prevent the healing of persistent air leaks. The pleural adhesions and areas of loculation may have prevented not only the proper functioning of the chest tubes but also the success of the chemical pleurodesis in both these patients.

SP occurs not infrequently as a complication of the lung disease in cystic fibrosis. Pneumothorax in cystic fibrosis is precipitated by the rupture of subpleural blebs or bullae and is associated with chronic inflammation, granulation tissue, and fibrosis rather than acute necrotizing pneumonias or abscesses.24,25 The lung diseases associated with cystic fibrosis and pediatric AIDS are quite different, but both are characterized by chronic inflammation and bacterial infections. In both of these diseases, pneumothoraces are difficult to treat and frequently recur.

Surgical intervention was the more efficacious treatment of the pneumothorax for the three children reported here. Pleurectomy and excision of intrathoracic cysts seems to be an aggressive approach in children with AIDS with a pneumothorax, but conservative management did not remove the source of the air leaks that predispose to pneumothorax in these three patients, nor did it address the problems of the chronic inflammation and loculations secondary to the pleural adhesions.

ACKNOWLEDGMENT: We wish to thank Dan Benevento for the photography of the pathologic specimens.

REFERENCES


CHEST / 108 / 4 / OCTOBER, 1995 1175
Use of Cardiopulmonary Bypass During Bronchoscopy Following Sand Aspiration*
A Case Report

James D. Mellema, MD; Susan L. Bratton, MD; Andrew Inglis, Jr., MD; and Jeffery P. Morray, MD

A 6-year-old boy with massive sand aspiration was effectively treated with femoral vein to femoral artery cardiopulmonary bypass (CPB), saline bronchial lavage, and exogenous surfactant. The patient was discharged the 9th hospital day without apparent sequelae. CPB should be considered for cases of sand or gravel aspiration when gas exchange is compromised.

(CHEST 1995; 108:1176-77)

CPB=cardiopulmonary bypass

Key words: aspiration; cardiopulmonary bypass; foreign body; surfactant

The following case represents the first reported use of cardiopulmonary bypass (CPB) in the treatment of sand aspiration.

Case Report

After aspirating an unknown quantity of sand, a previously healthy 6-year-old boy was noted to have a decreased level of consciousness and respiratory distress prompting tracheal intubation at the scene. Peak airway pressures (peak inspiratory pressure) of 80 to 100 cm H2O were required to achieve adequate chest rise. After transport to our emergency department, a chest radiograph was taken that demonstrated a "sand bronchogram" (Fig 1). Breath sounds were decreased bilaterally, and crepitus was noted over the thorax, abdomen, and scrotum. Because endotracheal suctioning resulted in severe desaturation, the child was transported to the operating room for bronchoscopy during femoral vein to femoral artery bypass. In spite of flow rates on bypass of 2.0 L/min/m2, the patient continued to have significant problems with oxygenation which only improved with an increase in peak inspiratory pressures from 50 to 80 cm H2O (Table 1). A rigid bronchoscope was inserted into the trachea, and during the subsequent 3 h gravel was removed from all segmental bronchi, and the bronchial tree was lavaged with normal saline solution. Following lavage, exogenous bovine surfactant (Survanta [beractant]), 2 mL/kg, was administered through the endotracheal tube. Following transfer to the pediatric ICU, the patient was ventilated with a high-frequency oscillator with a mean airway pressure of 25 cm H2O and a frequency of 5 Hz. The patient was converted to conventional mechanical ventilation on the 4th hospital day. He was extubated on the 7th hospital day and discharged on the 9th hospital day with no apparent neurologic or pulmonary sequelae.

Discussion

Ours is the first report of the use of CPB for treatment of sand aspiration. Previously, treatment of sand, gravel, or dirt aspiration has involved bronchoscopy and postural drainage without CPB,1-3 though CPB has been used for removal of other kinds of tracheal foreign bodies.4-6 In our patient, conventional methods of oxygenation and ventilation allowed adequate gas exchange only at very high peak inspiratory and mean airway pressures. Given the duration of bronchoscopy necessary and the degree of desaturation the patient experienced prior to CPB, it is unlikely that the patient would have survived without CPB. In spite of bypass

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*PEEP=positive end-expiratory pressure; PIP=peak inspiratory pressure.

*From the Department of Anesthesia and Critical Care (Drs. Mellema, Bratton, and Morray) and the Department of Otolaryngology (Dr. Inglis), Children’s Hospital and Medical Center, University of Washington School of Medicine, Seattle.