CASE RECORDS of the MASSACHUSETTS GENERAL HOSPITAL

Case 35-2024: A Newborn with Hypoxemia and a Lung Opacity

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CME

PRESENTATION OF CASE

Dr. Yanki Okuducu (Pediatrics): A newborn girl was admitted to the neonatal intensive care unit (NICU) of this hospital because of cardiorespiratory arrest at delivery.

The newborn's mother was a 19-year-old woman (gravida 1, para 1) with a history of hepatitis C virus infection. Pregnancy was complicated by urogenital *Chlamydia trachomatis* infection that was treated in the second trimester, as well as gestational hypertension. Serial ultrasound surveys of the fetal anatomy performed between 20 weeks 1 day of gestation and 39 weeks 6 days of gestation revealed dilatation of the urinary tract on the right side. Ultrasonography of the fetal heart was normal.

At 40 weeks 0 days of gestation, spontaneous rupture of membranes occurred, and the newborn's mother was admitted to this hospital. Fetal heart tracings indicated nonreassuring fetal status, and at 22 hours 39 minutes of labor, a cesarean section was performed. The uterus was incised, and the amniotic fluid was noted to be stained with meconium. Six minutes later, the newborn was delivered; she was in a breech position at delivery. The birth weight was 3575 g (62nd percentile), the length 50 cm (42nd percentile), and the head circumference 35.5 cm (72nd percentile).

The newborn had limp tone and no respiratory effort. Ventilatory support was initiated, and the trachea was intubated. Cardiopulmonary resuscitation was performed. Doses of epinephrine were administered through the endotracheal tube and an umbilical venous catheter, as were boluses of normal saline. After 16 minutes, spontaneous circulation was restored. The Apgar scores at 1, 5, 10, 15, and 20 minutes were 1, 0, 0, 1, and 3, respectively. The newborn was admitted to the NICU.

On the patient's arrival in the NICU, the axillary temperature was 34.9°C, the heart rate 141 beats per minute, and the blood pressure 84/59 mm Hg. The oxygen saturation was 97% while she was receiving oxygen through a mechanical ventilator in pressure-control mode. A course of therapeutic hypothermia was initiated.

Dr. Evan J. Zucker: Chest radiography (Fig. 1A) revealed a small-to-moderate pneumothorax in the medial aspect of the pleural cavity on the right side. Subtle lucency

in the middle-to-lower thorax was present on the left side, a finding attributed to a possible left pneumothorax or air trapping.

Dr. Okuducu: Needle thoracentesis was performed on the right side, and 20 ml of air was removed. Empirical treatment with ampicillin and ceftazidime was started. On day 2, increased work of breathing developed, and additional imaging was performed.

Dr. Zucker: Repeat chest radiography (Fig. 1B) showed resolution of the right pneumothorax and development of a large left pneumothorax with extensive collapse of the left lung. Needle thoracentesis was performed on the left side, and 27 ml of air was removed. Chest radiography performed after thoracentesis (Fig. 1C) showed that the left pneumothorax had decreased in size and inflation of the left lung had increased.

Dr. Angela R. Shih: Pathological examination of the placenta revealed meconium-stained membranes. There was evidence of acute chorioamnionitis with fetal vascular involvement and bacterial organisms on and in the membranes.

Dr. Okuducu: During the next 2 days, the difference between the preductal oxygen saturation and the postductal oxygen saturation ranged from 5 to 15 percentage points, a finding suggestive of persistent pulmonary hypertension of the newborn. The fraction of inspired oxygen on the mechanical ventilator was increased to 1.0, and the administration of inhaled nitric oxide was initiated. Hypotension was treated with fluid

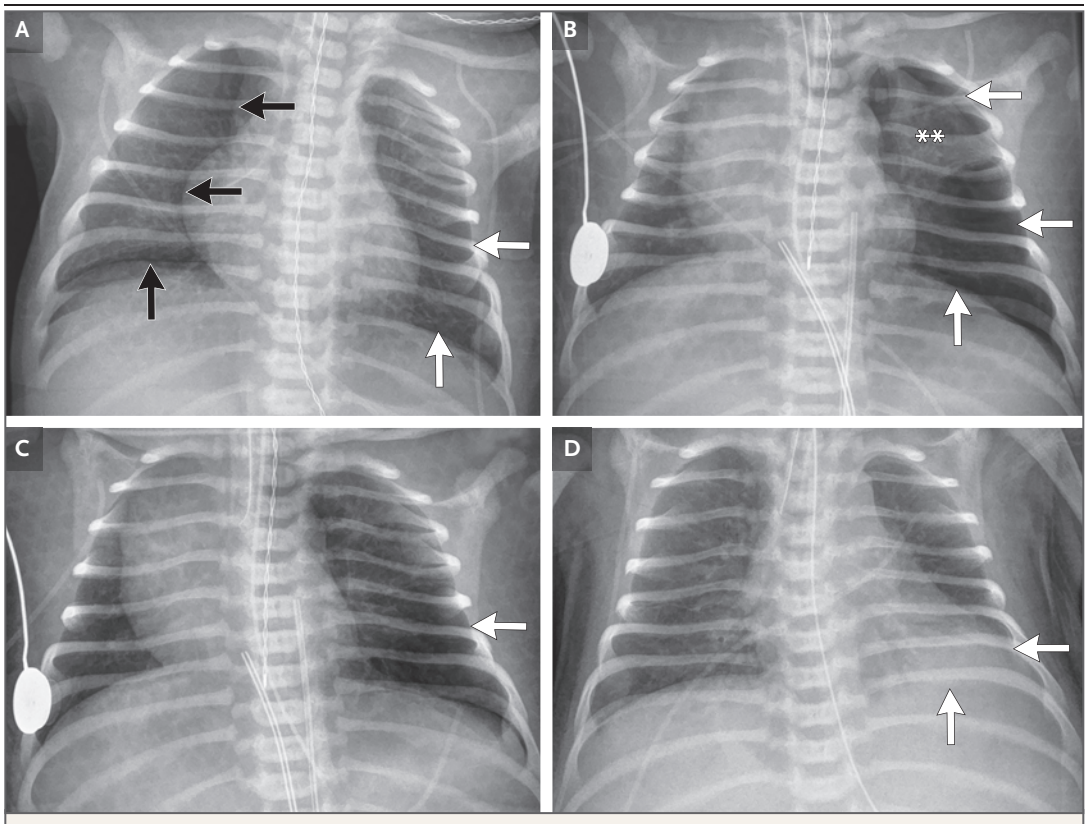

Figure 1. Chest Radiographs.

On the day of birth (Panel A), a small-to-moderate pneumothorax is present in the medial aspect of the pleural cavity on the right side (black arrows), and subtle lucency is present in the middle-to-lower thorax on the left side (white arrows), a finding attributed to a possible left pneumothorax or air trapping. On day 2, after needle thoracentesis is performed on the right side (Panel B), the right pneumothorax has resolved, and a large left pneumothorax has developed (arrows), with extensive collapse of the left lung (asterisks). After needle thoracentesis is performed on the left side (Panel C), the left pneumothorax has decreased in size, and inflation of the left lung has increased (arrow). On day 21 (Panel D), the pneumothorax findings have resolved, but an opacity is present in the left lower lobe (arrows) that has persisted since day 16, with silhouetting of the diaphragm and the border of the heart on the left side.

resuscitation, and dopamine and milrinone infusions were started. Empirical treatment with vancomycin was also started.

On transthoracic echocardiography, biventricular systolic function was at the low end of the normal range, and the right ventricle was moderately enlarged. A patent ductus arteriosus (measuring 2.9 mm in diameter), a midmuscular ventricular septal defect (measuring 2.0 mm in diameter), and a patent foramen ovale (measuring 2.5 mm in diameter) were present; each of these features was associated with bidirectional shunting. Tricuspid regurgitation (peak gradient, 58 mm Hg; arterial blood pressure, 66/36 mm Hg) and ventricular septal flattening were present, findings suggestive of elevated right ventricular pressure due to pulmonary hypertension.

Ultrasonography of the kidneys and bladder revealed severe dilatation of the urinary tract on the right side; the left kidney was normal. The patient had an episode of rhythmic myoclonic jerking of the left leg that progressed to shaking of the left leg and arm, and phenobarbital was administered. Electroencephalography confirmed that a seizure originating in the midline posterior region had occurred. The seizure had lasted 25 minutes and had resolved spontaneously before the administration of phenobarbital. Ultrasonography of the head was normal.

On day 4, the patient completed the 72-hour course of therapeutic hypothermia, and rewarming began. The blood pressure decreased. The milrinone infusion was gradually discontinued, the dopamine infusion dose was increased, an epinephrine infusion was started, and hydrocortisone was administered. The newborn had episodes of agitation that were associated with labile oxygen saturation. Chest radiography showed that the left pneumothorax had continued to decrease in size; patchy opacities were seen throughout the right lung. Transthoracic echocardiography showed depressed right ventricular function.

During the next week, the pressor dose was increased. The periods of decreased oxygen saturation became more frequent and more prolonged, and ventilatory support was increased. On day 11, the oxygen saturation was 95% while the patient was receiving oxygen through a mechanical ventilator in pressure-control mode (peak inspiratory pressure, 31 cm of water; positive end-expiratory pressure, 9 cm of water; fraction of inspired oxygen, 1.0), along with inhaled nitric oxide. Empirical therapy with vancomycin, ampicillin, and ceftazidime was stopped after the patient completed 10-day courses.

On day 16, the axillary temperature increased to 38.8°C. Chest radiography showed an opacity in the left lower lobe and a small left pleural effusion, as well as linear opacities in both upper lobes. Empirical treatment with oxacillin and gentamicin was started. On day 19, ventilatory support remained elevated, and empirical antibiotic therapy was transitioned to vancomycin and cefepime.

Dr. Zucker: On day 21, repeat chest radiography (Fig. 1D) showed a persistent opacity in the left lower lobe. Empirical therapy with vancomycin was discontinued.

Dr. Okuducu: A diagnostic test was performed.

DIFFERENTIAL DIAGNOSIS

Dr. T. Bernard Kinane: I participated in the care of this patient, and I am aware of the final diagnosis in this case. This full-term infant was delivered by cesarean section and underwent extensive resuscitation at birth. Chest radiography performed on the day of birth revealed a pneumothorax on the right side, as well as subtle lucency in the left lower lobe. The subsequent NICU course was complicated by multifactorial hypoxemic respiratory failure with severe persistent pulmonary hypertension. On day 16, chest radiography performed during an evaluation for new fever showed an opacity in the left lower lobe, in the area where lucency had been noted previously. A diagnosis of pneumonia was considered, and treatment with antibiotics was started, but the opacity persisted on chest radiography performed on day 21. In working toward identifying the diagnosis that is most likely to explain the patient's persistent parenchymal pulmonary opacity, I will begin by considering atypical presentations of two common conditions: meconium aspiration syndrome and congenital diaphragmatic hernia.

MECONIUM ASPIRATION SYNDROME

During the infant's delivery, the amniotic fluid was noted to be stained with meconium. In approximately 5% of infants with meconium-stained amniotic fluid, meconium aspiration syndrome develops; in 9.6% of patients with

meconium aspiration syndrome, a pneumothorax develops.[1,2] On chest radiography, meconium aspiration syndrome is characterized by diffuse pulmonary disease. In some cases, the disease may be isolated to a single lobe because of localized lobar bronchus occlusion. However, in the case of meconium aspiration syndrome, radiologic abnormalities are transient, and therefore, this diagnosis is unlikely to explain the persistent parenchymal pulmonary opacity seen in this patient on days 16 and 21.

CONGENITAL DIAPHRAGMATIC HERNIA

In children, when a lesion in the left thoracic cavity is observed on imaging, a congenital diaphragmatic hernia should be considered. Among the congenital lesions that can affect the lower lobe of the lung, congenital diaphragmatic hernia is by far the most common, with an incidence of approximately 2.4 cases per 10,000 live births.[3] The left thoracic cavity is involved in 80% of cases. The most common form of congenital diaphragmatic hernia is the Bochdalek hernia, which is a developmental defect in the posterolateral diaphragm that allows abdominal contents to herniate into the thorax.

In some — but not all — patients with a congenital diaphragmatic hernia, chest radiography shows bowel loops on the left side of chest, as well as rightward cardiomediastinal shift due to mass effect and hypoplasia of the left lung. Computed tomography (CT) of the chest can be performed to definitively rule out the diagnosis. This patient underwent serial chest radiography over the course of 3 weeks, and no findings consistent with a congenital diaphragmatic hernia were noted. These results make the diagnosis very unlikely, and therefore, I will consider other possibilities.

CONGENITAL MALFORMATIONS

It is important to note that the persistent parenchymal pulmonary opacity seen in this infant was located in the same area where subtle lucency had been observed on chest radiography performed on day 2. The persistence of radiographic changes in the same location strongly suggests that a congenital malformation in the lung was present at birth and later became infected, with the infection leading to fever and the persistent opacity on chest radiography. Congenital malformations of the lung include multiple bronchogenic cysts, congenital lobar overinflation, congenital pulmonary airway malformation (CPAM), and bronchopulmonary sequestration (BPS) (Fig. 2).[4]

Multiple Bronchogenic Cysts

Bronchogenic cysts form when bronchial tissue separates from the airway during development.[5] Such cysts are characteristically thin-walled and lined by ciliated pseudostratified columnar epithelium. Although bronchogenic cysts are predominantly found in the mediastinum, 5% of cases are found in the lung parenchyma, typically in the lower lobe of the lung. Multiple bronchogenic cysts can compress nearby bronchi, which may lead to obstruction and possibly pneumonia. However, bronchogenic cysts are usually large cysts that can be identified on prenatal ultrasound screening, so the diagnosis is unlikely in this case.

Congenital Lobar Overinflation

Congenital lobar overinflation is a condition that leads to overexpansion of a lobe of the lung.[6] The condition is an important consideration in this patient because overexpansion of the left lower lobe could explain the lucency seen on chest radiography performed on day 2. Congenital lobar overinflation is also associated with dyspnea and tachypnea, which were observed in this patient. However, involvement of the left upper lobe is most common, whereas involvement of a lower lobe is rare. In addition, patients with congenital lobar overinflation usually have extreme overexpansion, which would lead to more severe findings than those seen in this patient.

Congenital Pulmonary Airway Malformation

Bronchopulmonary malformations include CPAM and BPS. CPAM is caused by hamartomatous proliferation of airways during embryonic development and has an incidence of 1 case per 7500 live births. There are five types, each with distinctive clinical and morphologic features that correspond to their airway origins.[7] CPAM type 0 — now known as acinar dysplasia — is the least common type, accounting for 1 to 3% of cases. Acinar dysplasia can be ruled out in this patient because it is characterized by nonaerated lungs and is invariably fatal within the first day after birth.

CPAM type 1 is the most common type, accounting for more than two thirds of all cases, and it is often detected prenatally. Affected neonates typically have respiratory distress, which was seen in this patient. However, CPAM type 1 is unlikely in this patient because it is characterized

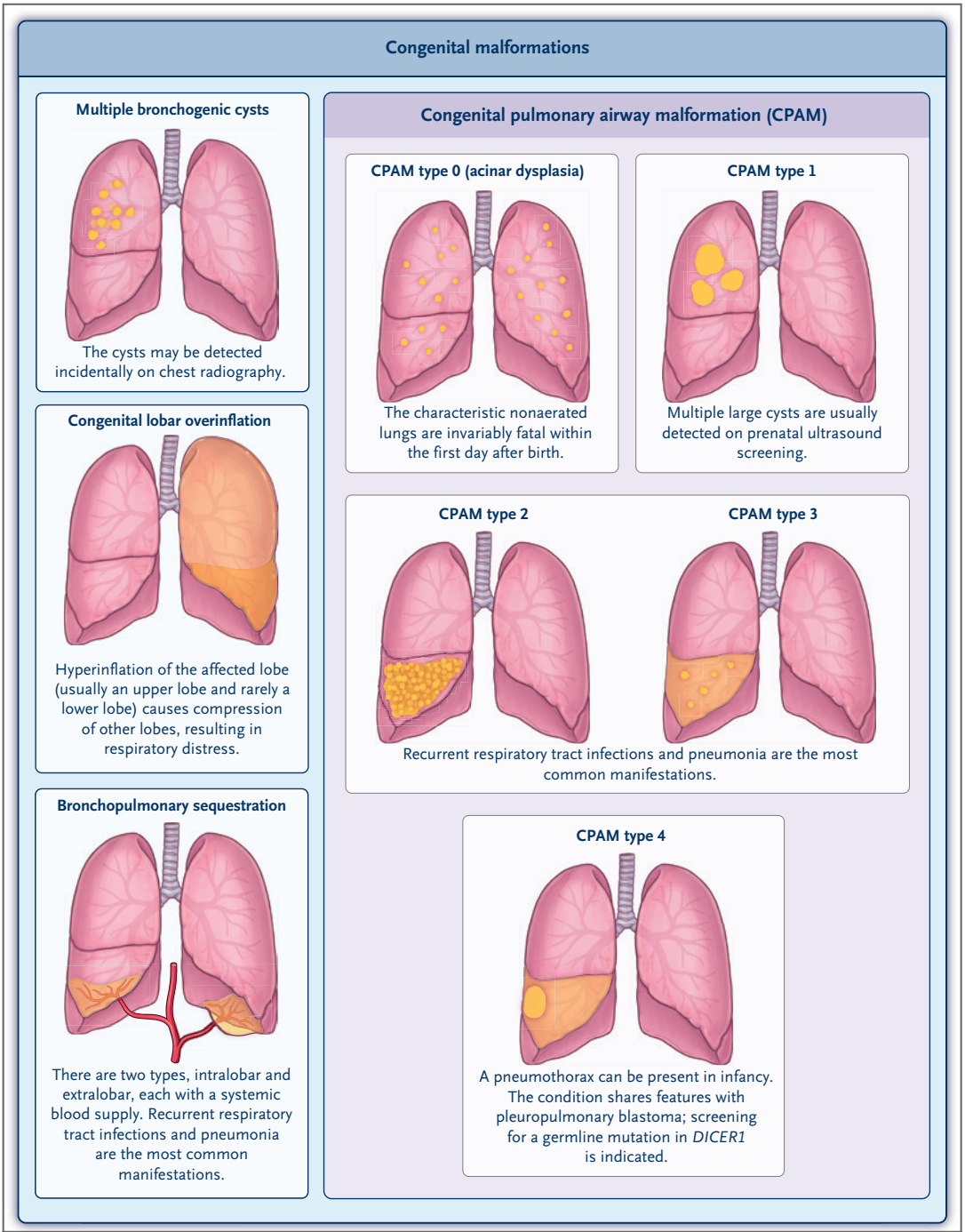

Figure 2. Congenital Malformations of the Lung.

The differential diagnosis for a congenital lesion in the left lower lobe includes multiple bronchogenic cysts, congenital lobar overinflation, congenital pulmonary airway malformation (types 0 through 4), and bronchopulmonary sequestration.

by large cysts (measuring up to 10 cm in diameter) that are usually detected on prenatal ultrasound screening and would also be easily identified on chest radiography.

CPAM type 2 is the second most common type, accounting for 10 to 15% of all cases. Affected patients present with respiratory symptoms within the first month after birth, and chest radiography shows multiple cystic spaces (measuring up to 2.5 cm in diameter) that result in a spongelike appearance. The findings on chest radiography in this patient were not consistent with CPAM type 2.

CPAM type 3 is less common, constituting 8% of all cases. Typically detected in utero or at birth, CPAM type 3 is unlikely in this patient because it is characterized by a solid, adenomatoid mass that contains cysts measuring up to 1.5 cm in diameter.

CPAM type 4 has clinical features that overlap with those of pleuropulmonary blastoma, a rare childhood lung tumor that may share origins with or emerge from CPAM type 4. Patients who present with relevant clinical features should be evaluated for a germline mutation in *DICER1*, which is seen in 40% of patients with pleuropulmonary blastoma and confers ongoing cancer risk for which future screening is indicated. CPAM type 4 is characterized by an acinar malformation lesion that consists of cysts of various sizes, all of which are lined by type 1 and 2 alveolar cells, with no mucous cells. Chest radiography usually shows peripheral cyst formation, but the cysts can be large enough to look like an overexpanded lobe. CPAM type 4 is also frequently associated with pneumothoraxes, which were seen in this patient. However, if the patient had CPAM type 4 with a superinfection, a cystlike structure would be identified on chest radiography. In addition, her pneumothoraxes have a more likely explanation: positive airway pressure associated with intubation and mechanical ventilation. Although another type of bronchopulmonary malformation is more likely to explain the patient's presentation, screening for a *DICER1* mutation would be prudent.

Bronchopulmonary Sequestration

BPS is a congenital malformation in which lung tissue is not connected to the tracheobronchial tree and the vascular supply to the involved lung parenchyma typically originates from the aorta.[8] The malformation can be categorized as extralobar or intralobar. Extralobar BPS is encapsulated by its own pleura and is typically situated between the lower lobe and the diaphragm. Intralobar BPS, which is more common than the extralobar form, is integrated into the normal lung parenchyma and is usually located in a lower lobe (in 98% of cases), particularly in the medial and posterior basal segments of the left lower lobe. On rare occasion, a hybrid lesion of CPAM and intralobar BPS can occur. Such hybrid lesions are predominantly observed in a lower lobe of the lung and can contain cysts of various sizes.

Although a hybrid of CPAM and BPS was possible in this case, I thought that intralobar BPS was the most likely explanation for the patient's persistent parenchymal pulmonary opacity because it is more common. I recommended ultrasonography of the chest as the first test to further evaluate for an aberrant vessel supply to confirm the diagnosis of intralobar BPS.

DR. T. BERNARD KINANE'S DIAGNOSIS

Bronchopulmonary malformation, most likely intralobar bronchopulmonary sequestration.

ADDITIONAL IMAGING STUDIES

Dr. Zucker: Ultrasonography of the chest with color Doppler (Fig. 3A) revealed a predominantly echogenic mass in the left lower lobe with a suspected arterial supply from the abdominal aorta. Confirmatory CT angiography of the chest (Fig. 3B), performed after the administration of intravenous contrast material, showed a heterogeneous masslike area of consolidation in the lingula and left lower lobe with a feeding artery arising from the abdominal aorta.

HOSPITAL COURSE

Dr. Katherine A. Sparger: The patient was evaluated by a pediatric surgeon, and the definitive management of her congenital bronchopulmonary malformation was deferred to the outpatient setting. Given her multiple congenital anomalies, the patient was seen for a genetics consultation. A chromosomal microarray showed no abnormalities, and an evaluation for a *DICER1* mutation was negative. The fever resolved, and

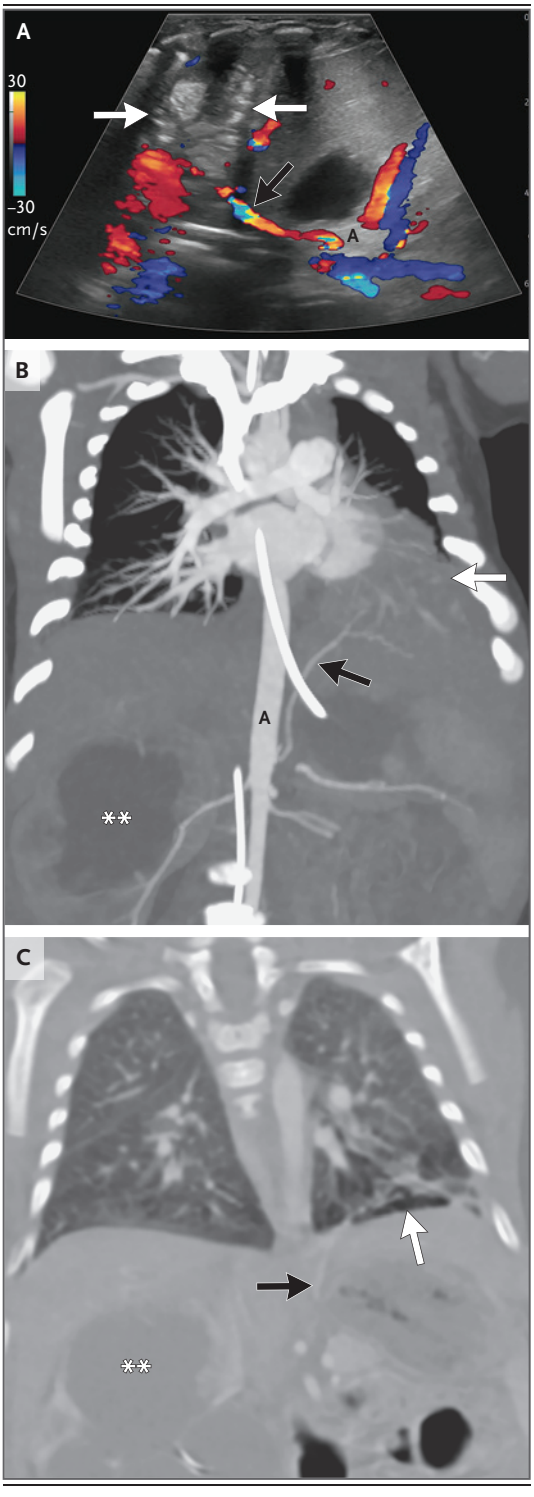

Figure 3. Additional Imaging Studies.

An ultrasound image of the chest with color Doppler obtained on day 21 (Panel A) shows a heterogeneous, predominantly echogenic mass (white arrows) centered in the left lower lobe with evidence of an arterial supply (black arrow) from the abdominal aorta (A). No pleural effusion is present. An oblique coronal maximum-intensity-projection image derived from contrast-enhanced CT angiography of the chest performed on day 23 (Panel B) confirms the presence of a heterogeneous masslike area of consolidation (white arrow) centered in the lingula and left lower lobe with a feeding artery (black arrow) arising from the abdominal aorta (A). The venous drainage could not be well delineated. Marked dilatation of the right renal collecting system (asterisks), a finding known from prenatal evaluation, is visible. A coronal image from follow-up CT angiography of the chest performed on day 149 (Panel C) shows that the previous area of consolidation has been replaced by hyperlucency (white arrow), a finding suggestive of cystic changes and air trapping; adjacent linear bands of probable atelectasis and scarring are present. The aberrant arterial supply (black arrow) to the affected lung from the abdominal aorta is present but less conspicuous, and the dilatation of the right renal collecting system (asterisks), now shown after pyeloplasty, is partially visible. On follow-up contrast-enhanced CT of the chest performed on day 183 (not shown), the findings were relatively unchanged.

all cultures remained negative. The courses of vancomycin and cefepime were completed. The infant was discharged home on day 43, after she was successfully weaned off all respiratory support and sedation and was feeding orally. She continued to receive care from a team of pulmonary, surgery, nephrology, and urology specialists.

DISCUSSION OF MANAGEMENT

Dr. Cassandra M. Kelleher: The patient was seen for a surgical consultation when the lesion in the left lower lobe was identified during her hospitalization. The presence of a feeding artery originating from the abdominal aorta to supply the lesion was strongly suggestive of BPS.[9]

Intralobar BPS is contained within the pleura of a lobe of the lung, most commonly the left lower lobe. Extralobar BPS is found outside the normal lung lobes and is enveloped by its own pleura. Intralobar BPS drains into the pulmonary veins, whereas extralobar BPS drains into the systemic circulation, typically the vena cava. In patients with BPS, the lung parenchyma is not connected to the tracheobronchial tree; however, intralobar BPS is connected to the alveoli of the normal lung by the pores of Kohn, through which bacterial contamination can occur, leading to infection.

At the surgical consultation, performed to consider management options for the patient, surgery was not indicated because of the increased risk associated with operating in an infected field and operating on a patient younger than 1 month of age. After the patient was discharged from the NICU, she was seen for planned surgical follow-up, and additional imaging was performed.

Dr. Zucker: Follow-up CT angiography of the chest, performed when the patient was approximately 5 months of age (Fig. 3C), showed that the previous area of consolidation had been largely replaced by hyperlucency suggestive of cystic changes and air trapping; adjacent linear bands of atelectasis and scarring were present. The feeding artery to this region of abnormal lung could still be seen but was less conspicuous.

Dr. Kelleher: The imaging findings were thought to indicate either intralobar BPS or the presence of a hybrid lesion containing components of both BPS and CPAM. Lobectomy is the standard treatment for both intralobar BPS and CPAM, given the risk of recurrent pneumonia involving the lesion. Infection occurs in up to 70% of patients with intralobar BPS, with symptoms of infection developing as early as 6 months of age.[9,10] Lobectomy for a congenital cystic lung lesion is often performed when the patient is between 4 and 6 months of age. In a patient younger than 1 month of age, the procedure poses an increased risk of respiratory complications; in a patient between 6 and 9 months of age, the procedure poses an increased risk of technical challenges related to scarring and lymphadenopathy from infection, even when the infection has been asymptomatic.[11] This patient's lobectomy was delayed because she underwent a more urgent procedure to relieve severe hydronephrosis that was unrelated to her lung lesion. The lobectomy was further delayed because of a series of febrile illnesses but was ultimately performed when she was 10 months of age.

Thoracoscopic resection is associated with a lower risk of long-term effects on pulmonary function than open thoracotomy and is therefore preferable when the surgery and anesthesia teams are comfortable and experienced with thoracoscopy in infants. A thoracoscopic lobectomy of the left lower lobe was planned for this patient. The initial thoracoscopic examination of the left hemithorax revealed a nearly obliterated pleural space, with dense adhesions in the area

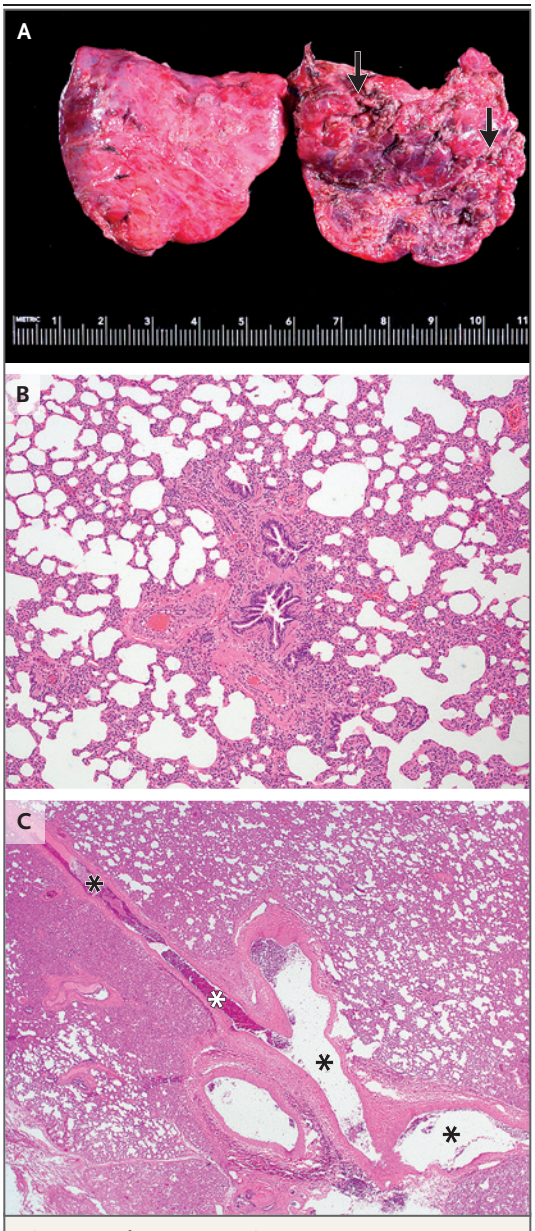

Figure 4. Lobectomy Specimen.

The resected left lower lobe shows an area of ragged pleural adhesions (Panel A, arrows); no evidence of a systemic feeding vessel is visible on gross examination. Hematoxylin and eosin staining shows nonspecific interstitial cellularity along with normal bronchovascular bundles; no evidence of parenchymal cysts, bronchiolar cystic proliferations, or a primitive blastemal proliferation is shown (Panel B). A large unpaired artery is present (Panel C, asterisks), a finding consistent with a systemic feeding vessel. The overall findings are consistent with intralobar bronchopulmonary sequestration.

spanning from the lung to the chest wall and diaphragm and in the major fissure. Lysis of adhesions was undertaken to free the lower lobe, and the inferior pulmonary ligament was incised up to the inferior pulmonary vein; a systemic feeding vessel was not identified. Attempts to identify the anatomical structures within the major fissure were unsuccessful, and ultimately, the procedure was converted to a thoracotomy. The patient had no complications and was discharged home 5 days after the procedure.

PATHOLOGICAL DISCUSSION

Dr. Shih: Gross examination of the resection specimen revealed a partially disrupted left lower lobe, measuring 9.5 cm in diameter, with an area of ragged pleural adhesions (Fig. 4A). A systemic feeding vessel was not identified. Serial sections showed a vague area of parenchymal discoloration that was poorly defined.

Histologic examination revealed predominantly intact alveolar architecture throughout the lung parenchyma, with only scattered foci of mild, nonspecific interstitial cellularity (Fig. 4B). A single, large-caliber muscular artery that did not appear to be paired with a bronchiole was present, a finding consistent with a systemic feeding vessel (Fig. 4C), which had been identified on imaging. Otherwise, normal bronchovascular bundles that were paired with bronchioles and arterioles of a similar caliber were present. No evidence of parenchymal cysts, bronchiolar cystic proliferations, or a primitive blastemal proliferation was noted. The overall morphologic findings, in conjunction with the imaging findings, are consistent with intralobar BPS.

PATHOLOGICAL DIAGNOSIS

Benign congenital pulmonary lesion that is most consistent with intralobar bronchopulmonary sequestration.

FOLLOW-UP

Dr. Kinane: Two years after this admission, the patient continues to be seen for follow-up in the pediatric pulmonary clinic. She has no respiratory symptoms. Because she had multiple congenital malformations, we are continuing to monitor for symptoms that would be suggestive of an underlying syndrome. However, at this point, no syndrome has been identified. Because she had a severe illness that led to resuscitation at birth, we are closely monitoring her development. She has been reaching all developmental milestones, but she will continue to be monitored as she enters the school system.

FINAL DIAGNOSIS

Intralobar bronchopulmonary sequestration.

This case was presented at Pediatric Grand Rounds.

Disclosure forms provided by the authors are available with the full text of this article at NEJM.org.

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