Mycetoma Within an Intralobar Sequestration*

Evidence Supporting Acquired Origin for This Pulmonary Anomaly

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A rare example of an intralobar sequestration (ILS) containing a fungal mycetoma is reported. This finding indicates the presence of a communication between the ILS and the airways, thus supporting the theory that ILSs are acquired lesions, rather than congenital malformations.

(ILS = intralobar sequestration)

Sequestrations are rare anomalies in which local areas of lung receive systemic arterial supply and lack normal communication with the tracheobronchial tree. They may occur within the normal pleural covering of the lung (intralobar sequestration [ILS]) or external to the pleura surface (extralobar sequestration). While extralobar sequestrations are generally considered congenital in origin, there is considerable debate regarding the pathogenesis of ILS, and the theory that ILS is an acquired, rather than a congenital, defect has gained popularity. We report the rare occurrence of a fungal mycetoma within an ILS and discuss implications for the pathogenesis of this anomaly.

CASE REPORT

A 36-year-old white woman was referred to our institution because of an enlarging mass in the lower lobe of the left lung. Twelve months previously, she had experienced left back pain radiating to her left shoulder associated with dyspnea on exertion, easy fatigability, and fever. A chest radiograph showed a well-defined, homogeneous density in the left lower lobe adjacent to the descending aorta, and a computed tomographic scan localized the mass to the posterior basal segment of the left lower lobe. Bronchoscopy revealed normal endobronchial anatomy. No communications were seen between the bronchi and the mass. Angiography demonstrated a separate blood supply to the lesion from an abnormal artery arising from the inferior phrenic artery (off the celiac axis).

Thoracotomy was performed, revealing a large mass in the left lower lobe with a 3- to 4-mm-diameter feeding artery in the inferior pulmonary ligament originating below the diaphragm. The artery was ligated and divided, and a left lower lobectomy was performed. The patient experienced an uneventful recovery.

Pathologic Findings

The specimen consisted of a left lower lobe weighing 230 g and measuring 12.0 × 10.5 × 5.0 cm. The visceral pleura was thin and glistening except in a 5.0 × 5.0-cm area at the base, where it was thickened by fibrofatty adhesions. A 1.5-cm-long segment of thick-walled artery measuring 0.4 cm in diameter was noted to enter the lung in this area.

Upon sectioning the lung, a 6.0 × 5.0 × 5.0-cm white to yellow, sharply demarcated, multiloculated cystic area was found in the posterior basal segment (Fig 1). The component cysts ranged in diameter from 0.3 to 4.5 cm, and many contained thick white to yellow purulent-appearing material. Embedded within this material in some of the cysts, but not attached to the cyst walls, were multiple small, 0.5- to 1.0-cm light brown to green, soft spherical masses. The large artery noted on the pleura overlying the cystic area could be traced to the center of the lesion. No gross evidence of communication between the bronchial tree and the cystic lesion could be found. The remaining lung parenchyma away from the cystic lesion was normal.

On microscopic examination, the cysts were lined by pseudostratified, ciliated columnar respiratory tract epithelium that was focally ulcerated. There was a thin rim of interstitial fibrosis and chronic inflammation surrounding the cystic area. An exudate containing acute inflammatory cells was present in some cysts. The grossly noted green-brown spheres were small mycetomas composed of...
dendically packed fungal hyphae arranged in lamellae, morphologically resembling Aspergillus species (Fig 2). No evidence of fungal invasion into adjacent lung tissue was found.

DISCUSSION

Recurrent infections are common complications of ILS, and often are the first manifestations of this anomaly. They are usually caused by common bacteria, although tuberculosis and nocardiosis have been implicated occasionally. Most infections in ILS can be explained either by the presence of collateral ventilation between the airspaces of the ILS and adjacent lung parenchyma or by bloodstream seeding during bacteremia. Fungal mycetomas complicating ILS, as occurred in our patient, have been reported only rarely, and are more difficult to explain.

Mycetomas are aggregates of fungal hyphae, most often Aspergillus species, occurring within previously abnormal, usually cystic areas of lung. The airways are thought to be the source of infection, since Aspergillus commonly resides in these structures even in normal individuals. The occurrence of a fungal mycetoma within an ILS indicates that the ILS must have some connection with the tracheobronchial tree, and in fact, such communications have been documented by bronchography in a few cases. Their presence supports Stocker’s theory that ILSs are acquired lesions related to chronic infection, rather than being congenital abnormalities related to abnormal development of the lung bud (in which case connection with the airways would be absent). According to this theory, the systemic arterial supply of an ILS results from hypertrophy of normally occurring arteries in the inferior pulmonary ligament, and residual communication with the bronchial tree could occur depending on the extent and chronicity of the infection.

REFERENCES

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Adult Respiratory Distress Syndrome as a Complication of Postanginal Sepsis*

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Adult respiratory distress syndrome (ARDS) was described in 1971 by Petty and Ashbaugh. Since that time it has been reported in association with many disease entities. We report a case in which a patient with postanginal sepsis, also known as Lemierre’s syndrome, had development of ARDS.

ARDS = adult respiratory distress syndrome; PEEP = positive end-expiratory pressure

Postanginal sepsis is characterized by persistent pharyngitis, toxic appearance, Pseudomonas aeruginosa bacteremia, empyema, pneumatoceles, septic arthritis, negative pharyngeal cultures for group A Streptococcus, and other distant sites of infection. First described by Schottmuller in 1918, it was best characterized by Lemierre in 1936. In the preantibiotic era, the mortality rate was 90 percent; a recent review by Moreno et al indicates a 12 percent mortality rate in the modern era. To our knowledge, adult respiratory distress syndrome (ARDS) has not been reported previously as a complication of postanginal sepsis.

CASE REPORT

A 19-year-old man with no medical history presented to our institution with 1-week duration of malaise, fever, sore throat, and 2 days of trismus. He was noted to be febrile to 38.6°C, have anterior cervical adenopathy, and an exudative pharyngitis; otherwise, results of his examination were unremarkable. His white blood cell count was elevated to 16.4 × 10⁹/L and his monospot was negative. He was believed to have bacterial pharyngitis and was treated with oral erythromycin and discharged from the hospital. Over the next 2 days, his clinical condition deteriorated, with left

Figure 1. Computed tomographic scan of the neck demonstrating right parapharyngeal collection (arrow).