Bronchial Dehiscence Associated with a Large Broncholith in a Lung Transplant Recipient*

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A 63-year-old man who underwent single lung transplantation for advanced emphysema had a postoperative course complicated by asymptomatic bronchial dehiscence associated with a large broncholith. The stone eventually caused airway obstruction requiring partial fragmentation and incomplete extrication. We suggest that calcified nodes of significant size be removed at the time of surgery in the lung transplant recipient. (Chest 1992; 102:1273-74)

**CMV = cytomegalovirus; SLT = single lung transplantation**

Pulmonary broncholithiasis has been well-described and is associated with a number of complications, including hemoptysis, atelectasis, bronchial obstruction with recurrent pneumonia, and bronchial fistula formation. We now report a case of bronchial dehiscence following single lung transplantation (SLT) that was due, in part, to a large broncholith situated at the site of surgical Anastomosis.

CASE REPORT

The patient was a 63-year-old man who had undergone right SLT for severe emphysema (pretransplant FEV₁ of 0.61 L). He was discharged home 18 days after transplantation with markedly improved functional status and an increase in FEV₁ to 1.65 L. Four weeks after transplantation, when completely asymptomatic, the patient underwent routine inspection bronchoscopy and a large broncholith measuring approximately 3.0 x 1.5 cm was observed extruding into the right main-stem bronchus at the site of surgical anastomosis (Fig 1). The suture line had broken down, resulting in partial bronchial dehiscence. Computed tomography of the chest demonstrated penetration of the broncholith into the bronchial lumen, with focal bowing of the bronchus anteriorly, consistent with dehiscence. No other evidence of mediastinal abnormality was identified.

The patient was hospitalized for observation, and on the following day, the bacterial cultures of the bronchoalveolar lavage fluid grew Klebsiella; appropriate antibiotic therapy was begun. A rigid bronchoscopic inspection was performed on hospital day 7 and the broncholith was judged too large for transbronchoscopic removal.

In addition, the treating physicians were concerned about the potential for inducing a severe mediastinitis. Instead, the broncholith was partially fragmented using large alligator forceps via the rigid bronchoscope, followed by extrication of the fragments. The patient’s subsequent hospital course was complicated by cytomegalovirus (CMV) pneumonitis, managed successfully with ganciclovir. The exact role of CMV in the dehiscence is not clear.

During flexible bronchoscopic inspection on hospital day 25, the stone and unstable anterior bronchial wall were observed to be causing marked airway obstruction on expiration; concomitant spirometry revealed a decrease in FEV₁ to 0.43 L. Rather than subject the patient to further invasive procedures, and due to his relative lack of symptoms, we opted for conservative management with observation only. The patient was eventually discharged home feeling well on hospital day 33. Follow-up spirometry as an outpatient revealed a return of FEV₁ to 1.4 L. Subsequent bronchoscopy revealed further healing and significantly less obstruction.

DISCUSSION

Pulmonary hilar calcifications detectable on routine chest roentgenography are very common, largely due to dystrophic calcification in areas of inflammatory necrosis; previous infection with histoplasmosis and tuberculosis are most often implicated. ¹, ² Simple calcified nodes are benign; however, erosion through the bronchial wall may result in numerous symptoms. Persistent cough, hemoptysis, and recurrent pneumonia are reported most frequently. ¹, ², ³ Complications that have been associated with broncholithiasis include bronchoesophageal fistula, aortotracheal fistula, and erosion into the pleura. ⁴, ⁵

We are unaware of any previous report of bronchial dehiscence associated with broncholithiasis in a lung transplant recipient. The incidence of bronchial dehiscence following lung transplantation has been markedly reduced by the application of a bronchial wrap with a vascularized...
pedicle, of either omental or pericardial origin. Despite use of the latter "bronchial pericardiopy" technique, our patient developed a major bronchial dehiscence. It is unclear whether his anastomotic breakdown was primarily caused by irritation from the adjacent stone or occurred secondary to bacterial infection. Nevertheless, it is clear that his postoperative morbidity was increased by the presence of the broncholith.

Management of the large symptomatic broncholith is controversial. Spontaneous expectoration may occur with smaller stones, but it is less likely with stones as large as that encountered in our patient. Removal via the rigid bronchoscope has been advocated with relatively high success rates and is currently favored over the smaller flexible bronchoscope due to the limited capabilities of the latter instrument should hemorrhagic complications occur.

An alternative approach for broncholith removal uses the YAC laser; stone fragmentation is followed by conventional retrieval of the fragments.

Massive hemoptysis, recurrent pulmonary infection, fistula, and suspicion of carcinoma are indications for surgical intervention. Associated mediastinal scarring usually makes resective surgery technically difficult. Our patient had undergone very recent thoracotomy, which would have made a surgical approach quite difficult.

We opted for relatively conservative management, with partial stone fragmentation and removal via rigid bronchoscopy. Acceptable airway patency was restored, and the patient was able to avoid repeated thoracotomy with its attendant risks. At six-month follow-up, the patient had returned to full-time employment, with a stable respiratory status. To avoid the potential morbidity that can occur secondary to postoperative broncholithiasis, we recommend the removal of any calcified nodes of significant size during the lung transplantation procedure.

REFERENCES

6 Davis EW, Katz S, Peabody JW. Broncholithiasis, a neglected cause of bronchoesophageal fistula. JAMA 1956; 156:555-57
11 Brantigan CO. Endoscopy for broncholith. JAMA 1978; 240:1483

Pulmonary Abnormalities in Klippel-Trenaunay Syndrome

A Histologic, Ultrastructural, and Immunocytochemical Study

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Klippel-Trenaunay (KT) syndrome is a rare, sporadic, congenital vascular disease of unknown etiology. We describe pulmonary findings in an 18-year-old male patient followed up since birth with the KT syndrome. The patient developed pleural and pericardial serous effusions that led to an open lung biopsy. Previous pulmonary findings have been limited to thromboembolic phenomena and pulmonary vein varicosities. On the other hand, reports of lymphatic hyperplasia, aplasia, and hypoplasia in KT have been limited to the extremities. For the first time, we describe lymphatic involvement of the lung in KT. Theplexiform hyperplasia of the lymphatic channels with smooth muscle hyperplasia leading to lymphatic obstruction, pleural and pericardial effusions are new findings. The lymphatic nature of theplexiform channels was confirmed by immunohistochemistry. Von Willebrand factor and QD-END/10 monoclonal antibodies either did not react or reacted poorly with lymphatic endothelium, features used to distinguish lymphatic and venous endothelium. Ultrastructurally, the absence of basement membrane continuity further substantiated the lymphatic nature of the channels. From our findings, the lymphatic abnormality in the syndrome appears to be more generalized than previously thought. This entity should be distinguished from lymphangioleiomyomatosis to which it bears a superficial morphologic appearance. (Chest 1992; 102:1274-77)

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KT = \text{Klippel-Trenaunay}; \text{LAM} = \text{lymphangioleiomyomatosis}; \text{MABs} = \text{monoclonal antibodies}
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Klippel-Trenaunay (KT) syndrome is a rare syndrome characterized by the triad of varicose veins, bony and soft tissue hypertrophy, and cutaneous hemangiomata. Since the first description of this syndrome in 1990 by Klippel and Trenaunay, approximately 140 cases have been published in the literature. The etiology remains unknown and the syndrome is sporadic in occurrence. In its classic form, limb hemihypertrophy is seen along with arteriovenous malformations. Other associated anomalies include arteriovenous fistulas, lymphangiomatous anomalies, polydactyly/syndactyly, telangiectasia, asymmetric facial hypertrophy, and pleural involvement. The usual patient does well but surgical intervention is sometimes necessary. The patient described in this case report did not have the classic syndrome, but he had a variant of it. He had isolated enlargement of the face with telangiectasia and polydactyly. He presented in January 1990 with pulmonary infiltrates, pleural effusions, and cardiomegaly. Special studies revealed the cardiomegaly

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