Chronic Lithoptysis With Multiple Bilateral Broncholiths*

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A case of chronic lithoptysis with unusual features was undiagnosed for 20 years. The presence of multiple broncholiths in different segments of both lungs, not previously described, was confirmed endoscopically. Areas of unusual mucosal punctate calcifications, not appreciated on concurrent high-resolution CT scan, also are described.

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Lithoptysis or the spitting of stones was first described by Aristotle in 300 BCE and confirmed by Galen and Aretaeus, as cited in Faber et al. In the 17th century, Schenk and Graftenburg vividly described lithoptysis in a series of 17 patients, as cited in Gordonson and Sargent.

Boerhaave, in 1744, reported the remarkable case of the famous botanist Veillantius who expectorated more than 400 broncholiths. The first reported case in modern medical literature was in 1885 by Peterson and the largest broncholith reported was 135 g. It has been rarely mentioned in recent medical literature. We describe a recent case of chronic recurrent lithoptysis with CT scan and bronchoscopic evidence of bilateral multiple broncholiths.

REPORT OF A CASE

A 63-year-old woman, who was a nonsmoker and had a history of non-insulin-dependent diabetes, hypothyroidism, and depression, presented with a lower respiratory tract infection. Serial radiographs of the chest revealed a persistent abnormality in the mid-zone of the right lung which on CT scan was consistent with a "right middle lobe syndrome" with bronchiectasis, multiple calcified lymph nodes, and scarring (Fig 1). She reported a recent positive purified protein derivative test but was exposed in childhood to her mother's active tuberculosis.

Physical examination disclosed no abnormalities. Flexible fiberoptic bronchoscopy revealed apparent broncholithiasis protruding into the bronchial lumen in multiple segments of both lungs. These were seen in virtually all segments of the middle and lower lobes of the right lung as well as in the upper lobe of the left lung (Fig 2, left). In addition to these larger broncholiths which corresponded to the CT findings of calcified lesions, there were several areas with fields of multiple gritty, smaller punctate apparent calcifications visualized on endoscopy only (Fig 2, right). It remains unclear as to whether these much smaller calcific protrusions represented primary broncholiths or fragments which broke off and embedded into the mucosa.

In retrospect, the patient admitted to multiple episodes of lithoptysis over the past 20 years. Initial disregard of these reports by physicians caring for this patient who had a psychiatric history, prompted her to omit any reference to the episodes on subsequent medical interviews. When shown a photograph of the stones, she recognized them as similar to the material she had previously expectorated.

DISCUSSION

Broncholithiasis can be defined as the presence of calcified material within the lumen of the tracheobronchial tree. A broncholith is composed of 85 to 90% calcium phosphate and 10 to 15% calcium carbonate. The etiology of stone formation remains controversial. The stones are most commonly believed to be the result of a postinflammatory healing process, in which a peribronchial lymph node expands into the airway. The local environment becomes alkaline which promotes the precipitation of calcium and phosphate. It also has been

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FIGURE 1. CT scan demonstrating multiple bilateral areas encroached by broncholithiasis.
speculated that they are the result of erosion through the bronchus of contiguous calcified granulomas secondary to the constant motion of the cardiorespiratory system. Some may also result from calcification of aspirated food or material embedded within the lumen of the bronchus. Finally, they might represent luminal protrusion of calcified bronchial cartilage through the mucosa.9

Broncholithiasis has been associated with numerous infections with tuberculosis predominating in Europe and histoplasmosis in the United States.1,10 It also has been reported rarely with actinomycosis, cryptococcosis, nocardiosis, aspergillosis, coccidioidomycosis, and silicosis.11-14 In one series, a specific organism was rarely identified (2 out of 42) in surgically resected broncholiths.1 Another surgical series reported up to 36% of resected broncholiths with identifiable organisms.14 Broncholithiasis occurs with equal frequency in men and women at all ages but most frequently in the fifth and sixth decades of life.15 The broncholiths predominate, as in our patients, in the right bronchial tree possibly correlating with the normal anatomy of lymph node distribution.1,14 The presentation usually is nonspecific with a chronic nonproductive cough occasionally with hemoptysis, purulence, and postobstructive pneumonia. Rarely there are reports of lithotyrosis or “gritty” or sandy sputum.8 Complications of broncholithiasis range from hemoptysis, to recurrent pneumonia, to extremely rare bronchoesophageal, aortoesophageal, aortotraheal, or pleural fistulas.7,16,17 In one surgical study, 4% of all cases with broncholiths were found to have a concomitant bronchogenic carcinoma.9 Diagnostic radiographic criteria as summarized by Vix15 include a calcified focus that (1) disappears on repeat films, (2) changes position, and (3) results in signs of bronchial obstruction. CT scanning is superior in visualizing the calcified focus and its relationship to the bronchus.19

The management of broncholithiasis depends on the location, size, and secondary effects of the broncholiths. The overall prognosis is favorable, and in most cases observation and conservative therapy are sufficient. Attempted removal of broncholiths via flexible fiberoptic bronchoscopy has had limited success.14,20 Broncholith removal by rigid bronchoscopy has been reported with success rates of up to 87% with, however, the potential complications of severe intrabronchial bleeding.21 Hence, a surgical approach appears the most prudent in cases of severe hemoptysis, traction diverticula, carcinoma, recurrent pneumonia, or fistula formation. In a review by Faber et al,1 segmental resection was the favored surgical approach. In a patient who requires surgery, the most conservative approach is desirable. In a series by Cole et al.,14 surgical removal of the broncholiths alone with bronchoplasty was sufficient in 12% (three patients); treatment for the remaining patients included segmental resection and lobectomy.

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