Congenital bronchial cyst with recurrent submassive hemoptysis

Samir Nusair, MD,^a Shaden Salameh-Giryas, MD,^b Chaim Springer, MD,^a and Uzi Izhar, MD,^c Jerusalem, Israel

Congenital pulmonary parenchymal bronchogenic cysts are mostly diagnosed during adulthood.¹ Children are often asymptomatic or might present with symptoms related to compression of adjacent organs, such as cough, dyspnea, or dysphagia. However, adults more often present with symptoms of infection, such as suppurative bronchitis and pneumonia or hemoptysis. We present a patient with a pulmonary parenchymal congenital cyst who presented with significant hemoptysis, but the cyst was not removed at the time. Seven years later, he had recurrent submassive hemoptysis, for which he underwent resection of the cystic lesion.

Clinical Summary

A 47-year-old nonsmoking man presented with hemoptysis during the last 4 days (30-50 mL/d blood). Seven years earlier, he experienced a similar episode of hemoptysis. Chest computed tomography (CT) then revealed areas of alveolar infiltrates and consolidation in the right upper lobe consistent with pulmonary hemorrhage. Repeat chest CT performed several weeks afterward showed resolution of the parenchymal hemorrhage and a residual finding of a cystic lesion with thin borders in the right upper lobe (Figure 1, A). No further intervention was offered at this point.

Chest plain x-ray film and CT performed on admission revealed interstitial and alveolar infiltrates with marked airspace disease within the right upper lobe consistent with alveolar hemorrhage (Figure 1, B) accompanied by a fluid-filled cyst. Flexible fiberoptic bronchoscopic examination showed blood within the posterior and apical segments of the right upper lobe, with no evidence of an endobronchial lesion. Transbronchial biopsy specimens showed alveolar hemorrhage, with no evidence of inflammatory or granulomatous lesions. Stain and culture results for bacteria, fungi, and mycobacteria were all negative.

A thoracotomy was performed, and the patient underwent segmentectomy of the right upper lobe. Histopathologic examination revealed a benign cyst, with hemorrhage involving the adjacent pulmonary parenchyma. The patient had a noncomplicated postoperative course and remains asymptomatic 1 year afterward.

Figure 1. Chest CT performed several months after the first episode of hemoptysis showing an intrapulmonary cyst in the posterior segment of the right upper lobe (A) and study performed during the present event of hemoptysis with interstitial and alveolar changes in the same area (B).

Comment

Congenital bronchogenic cysts originate from the foregut and develop within the cleavage between the respiratory tract and the digestive tract. Cysts forming early in the embryonic life are located in the mediastinum. However, when they occur later during bronchial budding and branching, the cysts are formed within the lung.

Bronchogenic cysts are usually solitary, with a spherical shape and a thin wall. Usually these cysts contain serous fluid; however, if they become infected, they might communicate with the bronchi, accumulating air or forming an air-fluid level. The cysts are usually lined by pseudostratified ciliated epithelium; however, malignant transformation within these lesions has been reported, such as squamous cell carcinoma in adulthood^{2,3} and rhabdomyosarcoma in infancy.⁴

Most of the congenital cysts in adults are asymptomatic; however, hemoptysis is the most common presentation in symptomatic patients.¹ Hemoptysis is usually related to an infection. Other

From The Institute of Pulmonology,^a The Department of Internal Medicine (Mt Scopus),^b and The Department of Cardiothoracic Surgery,^c Hadassah University Hospital, and the Hebrew University–Hadassah School of Medicine, Jerusalem, Israel.

Received for publication June 20, 2002; accepted for publication Aug 6, 2002.

Address for reprints: Samir Nusair, MD, Institute of Pulmonology, Hadassah University Hospital, PO Box 12072, Jerusalem, Israel, 91120 (E-mail: samjack@shani.net).

J Thorac Cardiovasc Surg 2003;125:972-3

Copyright © 2003 by The American Association for Thoracic Surgery

0022-5223/2003 \$30.00+0

doi:10.1067/mtc.2003.34

complications of bronchogenic cysts include bronchitis, cough, dyspnea, and pain.¹ Hemoptysis occurring in association with congenital cysts is often significant and requires surgical removal of the cysts to gain long-term control of the bleeding.^{1,5}

In conclusion, congenital pulmonary cyst might present only in adulthood and be symptomatic. However, hemoptysis, concern about potential malignant transformation, or both argue for surgical removal of these cysts once detected. Although our patient was asymptomatic for several years after the first event of hemoptysis, he had recurrent submassive hemoptysis, which could have been avoided if the cyst was resected on the first symptomatic presentation.

References

1. Ribet ME, Copin MC, Gosselin BH. Bronchogenic cysts of the lung. *Ann Thorac Surg.* 1996;61:1636-40.
2. Svennevig JL, Bugge-Asperheim B, Boye NP. Carcinoma arising in a lung cyst. *Scand J Thorac Cardiovasc Surg.* 1979;13:153-5.
3. Cuypers P, De Leyn P, Cappelle L, Verougstraete L, Demedts M, Deneffe G. Bronchogenic cysts: a review of 20 cases. *Eur J Cardiothorac Surg.* 1996;10:393-6.
4. Murphy JJ, Blair GK, Fraser GC, Ashmore PG, LeBlanc JG, Sett SS, et al. Rhabdomyosarcoma arising within congenital pulmonary cysts: report of three cases. *Pediatr Surg.* 1992;27:1364-7.
5. Lardinois D, Gugger M, Ris HB. Bronchogenic cyst of the left lower lobe associated with severe hemoptysis. *Eur J Cardiothorac Surg.* 1999;16:382-3.