

Late Massive Hemoptysis after Transbronchial Biopsy of Hamartoma: An Involvement of Pulmonary Artery and Vein

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Pulmonary hamartoma is a common benign neoplasm that is usually asymptomatic and that arises in the periphery of the lung. However, when the tumor is growing without showing characteristic findings of hamartoma, such as involving calcification, fat density, and chondromatous contents in chest X-rays or computed tomography, the diagnosis is sometimes that problematic and definitive histological diagnosis should be established. We herein report a case with a massive hemoptysis 10 days after a successful transbronchial biopsy. A 69-year-old man who underwent mitral valve plasty 6 years earlier presented a left lung shadow during a routine chest X-ray. The shadow was seen to be growing by a series of chest X-rays. A week after warfarin had been stopped, a bronchoscopic biopsy was performed. No bronchial hemorrhage was observed during the procedure, and warfarin was not restarted. The patient began noticing bloody sputa once or twice a day, and 10 days after the biopsy, 400 mL of hemoptysis was suddenly disgorged. An emergency left upper lobectomy of the lung was performed, and the hemoptysis soon disappeared postoperatively. The patient is well without respiratory symptoms 36 months after the surgery. (Ann Thorac Cardiovasc Surg 2007; 13: 400–402)

Key words: hamartoma, bronchoscopy, complication, surgery, hemoptysis

Introduction

Pulmonary hamartoma is a common benign neoplasm and is usually asymptomatic. However, sometimes it is clinically problematic when its radiological appearance is mimicking primary lung cancer and/or when it grows rapidly. In such cases, a bronchoscopic biopsy is often attempted to define the histological diagnosis. Although a bronchial hemorrhage is one of the severe complications of transbronchial biopsy, a late massive hemoptysis is rare.

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We herein report a case with a massive hemoptysis that occurred 10 days after a transbronchial biopsy of a suspected hamartoma. The involvement of thick pulmonary vessels in the tumor was demonstrated preoperatively by dynamic computed tomography (CT). The patient was then successfully treated immediately by surgical resection. We present the detailed clinical features of this case with a review of the literature.

Case Study

A 69-year-old man who underwent mitral valve plasty 6 years earlier presented a left lung shadow during a routine chest X-ray (Fig. 1). He was referred to the respiratory medicine group at our hospital, and chest CT demonstrated a round mass in the apical area of the left lower lobe (Fig. 2A). Initially, the mass was suspected as hamartoma; however, the shadow was growing, as seen by a

series of chest X-rays, and a histological diagnosis of the tumor was needed to exclude primary or metastatic malignancy. A week after warfarin had been stopped, a bronchoscopic biopsy was performed. No bronchial hemorrhage was observed during the procedure, and warfarin was not restarted. The patient later noticed bloody sputa once or twice a day. Ten days after the biopsy, 400 mL of hemoptysis was suddenly disgorged, and the patient was immediately transferred to our hospital. A dynamic CT revealed that the pulmonary tumor was surrounded by irregular infiltrative low density that was due to acute hemorrhaging from the tumor, involving branches of pulmonary artery (Fig. 2B) and veins (Fig. 2C). Especially, the pulmonary artery branches were surrounding the tumor. An emergency left upper lobectomy of the lung was then performed, and the hemoptysis soon disappeared postoperatively. The specimen showed an involvement of the pulmonary artery and veins (Fig. 3). Histologically, a diagnosis of parenchymal hamartoma was made. The patient was discharged uneventfully 3 weeks later and is well without respiratory symptoms 36 months after the surgery.

Discussion

Pulmonary hamartoma is a common benign tumor that usually has slow growing potential.¹⁾ It is often found in the periphery of the lung, rarely in the hilar region.²⁾ In a chest X-ray or in CT image, the tumor is usually demonstrated as a solitary, nonspecific, and round-shaped mass. When fat density, calcification, and/or chondromatous content were demonstrated in the lesion, because these findings are thought to be strong evidence of hamartoma, the lung mass will then be observed by periodic chest X-rays or CT scans. However, when the lesion is growing without showing the characteristic findings of hamartoma, lung cancer should be excluded. A transthoracic needle biopsy is useful in the definitive diagnosis of hamartoma.³⁾ But in our case, the tumor existed in the hilar region; thus transbronchial biopsy was selected instead. If the biopsy had failed to define diagnosis, surgical intervention would be considered.

Hansen and co-workers reported the clinical features of 89 patients of pulmonary hamartoma that had been histologically proven by transthoracic biopsy or surgically resected specimens.¹⁾ Eight cases of the 89 were incidentally discovered and resected during thoracotomy for other thoracic diseases. Of the remaining 81 cases, the diagnoses were obtained only by transthoracic biopsy in 34 cases. A thoracotomy was followed after nondiag-



Fig. 1. The chest X-ray demonstrates a round, well-demarcated mass lesion in the left hilum. An artificial mechanical valve and sternal wires are also seen.

nostic transthoracic biopsies in 6 cases, and exploratory thoracotomies were performed without attempting transthoracic biopsies in 41 cases. We find from these results that more than half of pulmonary hamartoma cases need surgical intervention for a definitive diagnosis.

Transbronchial biopsy by fiber-optic bronchoscopy is also used in the diagnosis of pulmonary hamartoma.⁴⁾ However, we have found no reports on a sizable study of its clinical value in the diagnosis of pulmonary hamartoma in English literature.

The occurrence of newly developed or exacerbated hemoptysis after transbronchial biopsy via fiber-optic bronchoscopy was estimated 24.6% of the time and hemorrhaging of more than 25 mL 17.5%.⁵⁾ In other reports, bleeding of more than 20 mL during transbronchial biopsy was reported as 7.7%.⁶⁾ Pulmonary hamartoma is usually asymptomatic. Late massive hemoptysis, i.e., 10 days after the biopsy, as in our case, was not reported in the literature.

Kanauchi and associates showed 53% ($n=19$) of pulmonary hamartomas to have pulmonary artery involvement by thin section CT in Japanese literature.⁷⁾ They also demonstrated no involvement of the pulmonary vein. A similar study analyzing vascular involvement of hamartoma could not be found in English literature. In our case, both the pulmonary artery and vein were shown by dynamic CT to be involved in the tumor. Probably the inflammatory degeneration of the biopsied lung parenchyma near the tumor was advanced 10 days after the biopsy, resulting in a breakdown of some of these involved vessels, provoking a delayed massive hemoptysis. However,

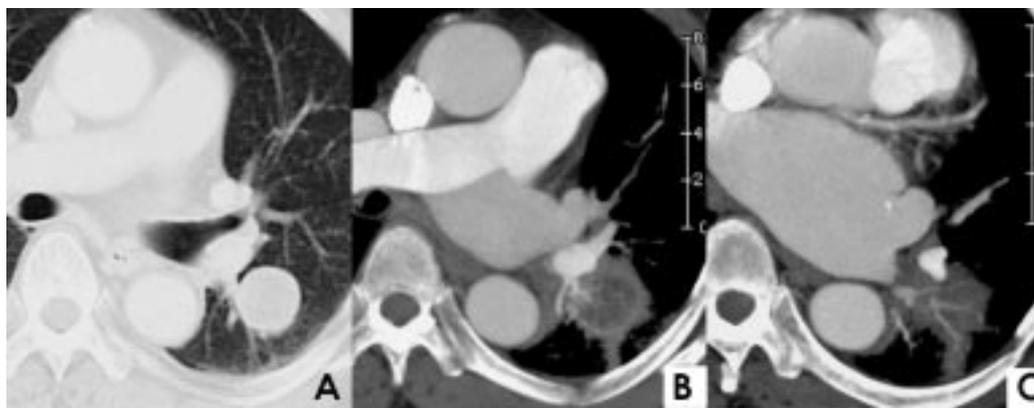


Fig. 2.

- A:** Chest computed tomography (CT) reveals the same smooth, round-shaped, and well-demarcated lung mass in the hilum of the apicoposterior segment of the left lower lobe.
- B:** A dynamic CT demonstrates an involvement of the pulmonary artery branches. Secondary low-density changes because of tumor bleeding surrounding the tumor and pulmonary artery branches are also visualized.
- C:** Dynamic CT also shows an involvement of pulmonary vein branches and surrounding infiltrative density by hemorrhaging.

this implication could not be proven by a histological examination of the specimen.

Transbronchial fiber-optic bronchoscopy is a safe method to obtain the histological diagnosis of pulmonary nodular lesions. Though when hamartoma is suspected, especially in such hilar cases, pulmonary vascular involvement and massive bronchial hemorrhaging even in a late phase should be considered, at which time careful bronchoscopic procedures and observations are needed.

References

1. Hansen CP, Holtveg H, Francis D, et al. Pulmonary hamartoma. *J Thorac Cardiovasc Surg* 1992; **104**: 674–8.
2. Fudge TL, Ochsner JL, Mills NL. Clinical spectrum of pulmonary hamartomas. *Ann Thorac Surg* 1980; **30**: 36–9.
3. Hamper UM, Khouri NF, Stitik FP, et al. Pulmonary hamartoma: diagnosis by transthoracic needle-aspiration biopsy. *Radiology* 1985; **155**: 15–8.
4. Panos RJ, Chang SW. Diagnosis of pulmonary hamartoma by bronchoscopic transbronchial biopsy. *J Natl Med Assoc* 1989; **81**: 460–3.
5. Diette GB, Wiener CM, White P, Jr. The higher risk of bleeding in lung transplant recipients from bronchoscopy is independent of traditional bleeding risks: results of a prospective cohort study. *Chest* 1999; **115**: 397–402.
6. Bjørtuft O, Brosstad F, Boe J. Bronchoscopy with transbronchial biopsies: measurement of bleeding vol-

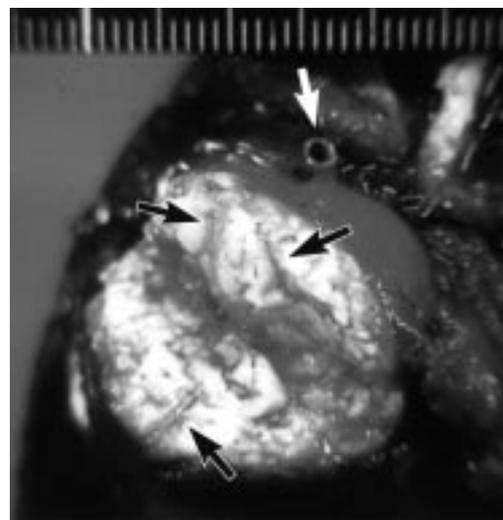


Fig. 3. The cut surface of the gross specimen reveals pulmonary artery involvement in the periphery of the tumor (white arrow). Small vessels were also seen in the cut surface of the tumor tissue (black arrows).

- ume and evaluation of the predictive value of coagulation tests. *Eur Respir J* 1998; **12**: 1025–7.
7. Kanauchi T, Hoshi T, Kato A. CT findings of pulmonary hamartoma with special reference to epithelial-lined clefts and connection with pulmonary arteries. *Nippon Igaku Hoshasen Gakkai Zasshi* 2004; **64**: 300–4.