Peribronchiolar Metaplasia of the Lung Incidentally Detected During a Surgery for Pneumothorax: A Case Report

Takehiro TSUCHIYA[*1], Naohiro IJIRI[*1], Kiko TOKUNAGA[*2], Satomi MIZUTANI[*2] and Tsutomu FUKUDA[*2]

[*1]*Department of Thoracic Surgery, Chigasaki Municipal Hospital*
[*2]*Department of Respiratory Medicine, Chigasaki Municipal Hospital*

(Received February 14, 2022; Accepted April 22, 2022)

Background: Peribronchiolar metaplasia (PBM) is a lesion characterized by an abnormal connection between the terminal bronchiole and parabronchiole via the Lambert's canals. We report a rare case of PBM incidentally detected during a surgery for pneumothorax.
Case presentation: At 38-year-old man was admitted to our hospital with chest pain. He was diagnosed with pneumothorax and treated using a thoracic drain 12 years ago. Chest computed tomography revealed a cyst in the right upper lobe and ground glass lesion with a solid component in the right lower lobe of the lung. Hence, we performed a surgery for pneumothorax management, which revealed a cyst in the right upper lobe and induration with angiogenesis in the right lower lobe. We performed partial resection of the right upper and lower lobes. Pathological examination of the lower lobe nodule revealed small airways with lymphocytic inflammation and bronchiolar metaplasia. Pathological diagnosis of the nodule was PBM.
Conclusion: Although PBM is considered a lesion with good prognosis, there have been cases associated with early-stage lung cancer. Hence, care should be taken to distinguish PBM from other neoplasms. However, preoperative diagnosis is difficult in most cases so complete surgical resection is recommended, if feasible.

Key words: Peribronchiolar metaplasia, Lambertosis, Ground glass lesion, pneumothorax surgery

INTRODUCTION

PBM is a lesion characterized by an abnormal connection between the terminal bronchiole and parabronchiole (Lambert's canals) [1]. It involves non-specific reactions around the Lambert's canals that result in the extension of bronchiolar type epithelial cells along the alveolar walls adjacent to the airways [2] in a process is called Lambertosis. PBM is a rare lung tumor that mimics early-stage lung cancer in X-ray images. In addition, there has been a report of PBM associated with early-stage lung cancer. Since preoperative diagnosis is difficult in most cases, complete surgical resection is recommended, if feasible. Herein we report a rare case of PBM that was incidentally detected during a surgery for pneumothorax.

CASE PRESENTATION

A 38-year-old man was admitted to our hospital with chest pain. He was diagnosed with right pneumothorax and treated using a thoracic drain 12 years ago. A chest X-ray showed that the right lung had collapsed. Hence, his condition was diagnosed to be a recurrence of the right pneumothorax (Fig. 1A). He was a medical care worker, with a smoking history of 5 cigarettes per day from 20 to 30 years old and had no history of occupational or environmental exposure or dust inhalation. Chest computed tomography (CT) revealed a cyst in the right upper lobe (Fig. 1B) and ground glass lesion with a solid component in the right lower lobe (Fig. 1C) of the lung. As the right lung was collapsed, we considered the nodule to be an inflammatory change, and thus, performed a surgery for pneumothorax management. Intraoperative findings revealed a cyst in the right upper lobe (Fig. 2A) and induration with angiogenesis in the right lower lobe (Fig. 2B) of the lung. We considered the lower lobe lesion to be a nodule, and hence, performed partial resection of right upper and lower lobes. The nodule in the right lower lobe measured 15 x 15 mm. The cut surface of the lesion had a firm-to-hard texture with tan-colored tissue (Fig. 3A). Pathological examination revealed small airways with lymphocytic inflammation and adenomatous hyperplasia (Fig. 3B, 3C). Dusty materials were contained in the small airways (Fig. 3D). The pathological diagnosis of the right lower lobe nodule was PBM. The patient remained free of recurrence for 1 year after surgery.

DISCUSSION

Lambert's canals are tubular connections between the terminal and respiratory bronchioles and adjacent alveoli, serving as accessory air channels of the distant alveoli by bypassing the main conductive airways [1]. Non-specific reactions around the Lambert's canals result in the extension of bronchiolar type epithelial cells along the alveolar walls adjacent to the airways, which is called Lambertosis [3]. PBM is a common finding in

Takehiro TSUCHIYA, Department of Thoracic Surgery, Chigasaki Municipal Hospital, Honson 5-15-1, Chigasaki, Kanagawa 253-0042, Japan
Tel: 81-467-52-1111 Fax: 81-467-54-0770 E-mail: tatsuchiya-ths@umin.ac.jp

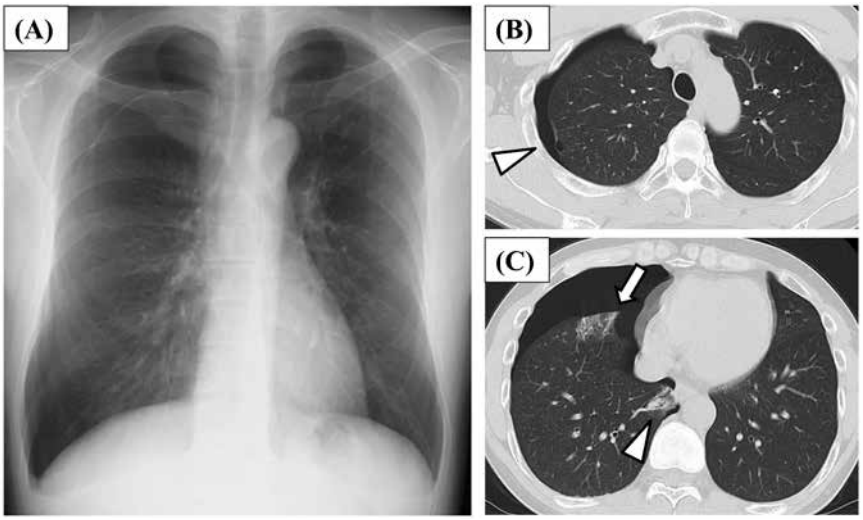

Fig. 1 A: Chest X-ray showing collapse of the right lung.
B: CT showing a cystic lesion in the right upper lobe (arrowhead).
C: CT showing ground glass lesion with a solid component in the right lower lobe (arrowhead). Another ground glass lesion disappeared after surgery (arrow).

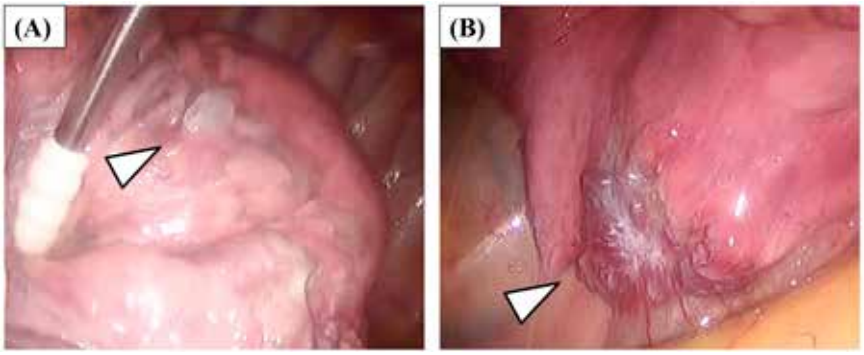

Fig. 2 Intraoperative findings
A: Lung cyst in the right upper lobe (arrowhead).
B: Tumor-like lesion with angiogenesis in the right lower lobe, which infiltrated the mediastinal pleura (arrowhead).

diffuse interstitial lung disease [4], patients are usually asymptomatic, and the lesions are often an incidental radiographic finding. The frequency and etiology of PBM are unclear; nevertheless, they are reported to be more common in women than in men [4]. However, since the CT showed no interstitial lung fibrosis, we considered the lesion to be a solitary one. In certain cases, a given pathologic finding is unique to a specific condition, such as respiratory bronchiolitis in cigarette smokers [3]. This patient also had a smoking history of 5 cigarettes per day for a decade. In previous cases CT has shown ground glass nodules, and in some instances a solid component had appeared in the process [5]. The potential differential diagnoses for PBM include atypical adenomatous hyperplasia, adenocarcinoma in situ, microinvasive adenocarcinoma, and mycobacteriosis [3, 6]. Additionally, one case reported that positron emission tomography (PET) with 2-[18F] fluoro-2-deoxy-D-glucose showed no abnormal uptake in the nodule [5]. Hence, clinical usefulness of PET with 2-[18F] fluoro-2-deoxy-D-glucose in PBM is unclear. Our case also showed ground glass nodules with a solid component in the right lower lobe. However, as the right lung was collapsed, we considered the nodule to be an inflammatory change. In fact, another ground glass lesion of the right lower lobe disappeared after surgery (Fig. 1C). The pathological features of PBM include bronchiolar metaplasia, goblet-cell metaplasia, or squamous metaplasia of the surface lining of adjacent scarred alveolar septa [7]. Hence, it is thought that image findings of ground glass lesions are derived from thickening of the alveolar wall and a solid component is derived from fibrous hyperplasia in the stroma. Our pathological analysis showed that dusty materials were contained in the small airways. We also detected induration with angiogenesis in the right lower lobe during the operation. Therefore, we considered that PBM could cause solid component due to the small partial obstructive pneumonia. In addition, ground glass lesions were derived from adenomatous hyperplasia along the surface of the alveolar wall. Although PBM is considered a lesion with good prognosis, care should be taken to distinguish it from other neoplasms because a previous report described a nodule which was histologically diagnosed as PBM with a co-existing adenocarcinoma in situ within the nodule [8]. In our case, as postoperative pathology didn't reveal the lung cancer, we thought that we could completely resect the

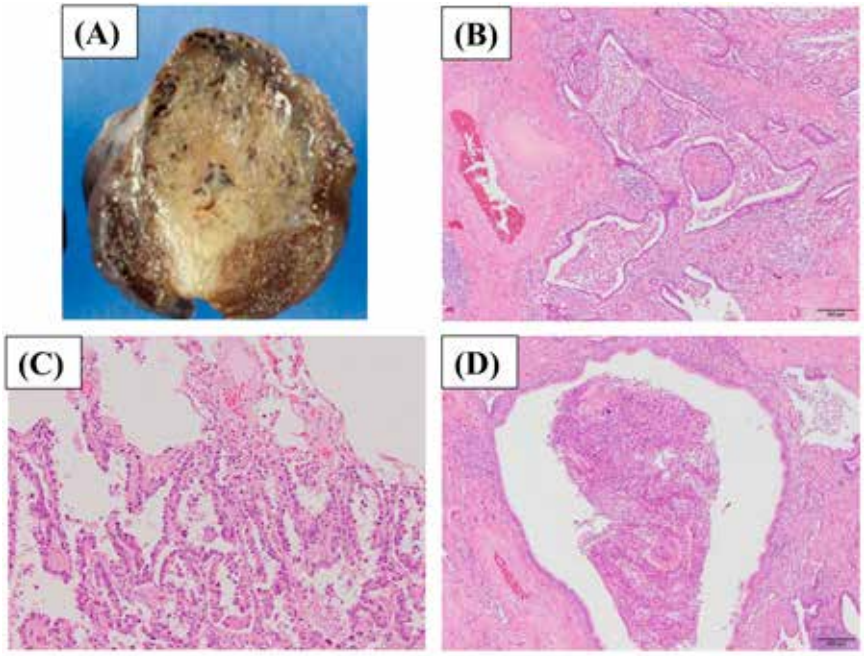

Fig. 3 Resected specimens of the tumor-like nodule from the right lower lobe
A: The nodule was 65 x 30 x 20 mm. The cut surfaces showed firm-to-hard, tan-colored tissue.
B: Pathological examination revealed small airways with lymphocytic inflammation. Original magnification x 4.
C: Pathological examination revealed adenomatous hyperplasia along the surface of the alveolar wall. Original magnification x 10.
D: Dusty materials were contained in the small airways. Original magnification x 4.

lesion. If the lesion was lung cancer, we had considered to perform the right lower lobectomy. However, preoperative diagnosis is difficult in most cases; we incidentally detected the nodule during a surgery for pneumothorax in our case. Hence, complete surgical resection is recommended, if feasible, for accurate diagnosis. This would aid in early identification and treatment of malignant tumors.

ETHICS STATEMENT

The patient provided informed consent, and patient anonymity was preserved.

REFERENCES

1) Lambert MW. Accessory bronchiole-alveolar communications. J Pathol Bacteriol 1955; 70: 311–4.
2) Hasleton P, Flieder DB. Spencer's pathology of the lung, 6th ed. Cambridge: Cambridge University Press, 2013: 646.
3) Colby TV. Bronchiolitis: pathologic considerations. Am J Clin Patho 1998; 109: 101–9.
4) Fukuoka J, Franks TJ, Colby TV, Flaherty KR, Galvin JR, Hayden D, *et al*. Peribronchiolar Metaplasia: a common histologic lesion in diffuse lung disease and a rare cause of interstitial lung disease: clinicopathologic features of 15 cases. Am J Surg Pathol 2005; 29: 948–54.
5) Yasuura Y, Kayata H, Mizuno K, Miyata N, Kojima H, Isaka M, *et al*. Solitary peribronchiolar metaplasia showing a sub-solid nodule on computed tomography. General Thoracic and Cardiovascular Surgery 2019; 67: 1093–6.
6) Yanagawa N, Kato H, Kanauchi N, Oizumi H, SadahiroM, Motoyama T. Two cases of solitary peripheral small lung tumor needed to differentiate from small lung adenocarcinoma. Jpn J Diagn Pathol 2007; 24: 426–9.
7) Cagle PT, Allen TC, Beasley MB, Borczuk A, Butt YM, Dacic S, *et al*. Pulmonary pathology: An Atlas and Text, 3rd ed. Philadelphia: Wolters Kluwer, 2019: 329–30.
8) Suzuki Y, Saito Y, Odaka H, Kurokawa H, Enomoto K, Kawai H. A case of peribronchiolar metaplasia accompanied by adenocarcinoma in situ. Jpn J Chest Surg 2017; 31: 901–4.