A Case of Thoracolithiasis Extracted with a Thoracoscope

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We report on an 81-year-old male patient in whom chest computed tomography during follow-up for extramammary Paget's disease revealed a nodule in the right thoracic cavity. Because he had been taking methotrexate for rheumatoid arthritis, the possibility of methotrexate-associated lymphoproliferative lung disease was considered and methotrexate was discontinued as a precaution. No calcification was found inside the nodule, and there was no change in its size or position for 6 months. The patient had a history of malignant tumor, so thoracoscopic surgery was performed for diagnostic purposes. A free white nodule was found in the right thoracic cavity and was subsequently removed. The pathological diagnosis was thoracolithiasis; although the nodule had not moved since its discovery, movement was suspected to have occurred 3 years prior. Although thoracolithiasis is rare, it should be considered as a differential diagnosis for nodules at the end of the lung field and on the diaphragm.

Key words: pleural stone, thoracolithiasis, thoracoscopy

INTRODUCTION

Thoracolithiasis is a rare disease. If movement within the thoracic cavity is confirmed, this can aid a differential diagnosis before surgery; however, if movement cannot be confirmed, preoperative diagnosis is often difficult. We report a case of thoracolithiasis identified on chest computed tomography (CT) during follow-up for other diseases, which was removed under thoracoscopic surgery.

CASE REPORT

An 81-year-old male had extramammary Paget's disease (diagnosed at 76 years of age), left empyema thoracis (diagnosed at 78 years of age), and rheumatoid arthritis (diagnosed at 80 years of age). He had no history of smoking, and he had no unusual occupation or family history. In the 5th year after surgery for extramammary Paget's disease, a nodule in the right thoracic cavity was identified on chest CT (Fig. 1). CT 3 years prior had not identified any nodules at the same position. Positron emission tomography-CT was performed to determine the presence or absence of metastasis of extravascular Paget's disease, but no abnormal accumulation was observed. The patient had developed rheumatoid arthritis and received methotrexate therapy for 7 months before the nodule was identified. The methotrexate was discontinued given the possibility of methotrexate-associated lymphoproliferative lung disease, but the nodule had not changed at 6 months. In addition, although no nodule had been recognized at this site on CT 3 years prior, a shadow similar to that of the nodule was identified between the esophagus and the inferior vena cava in the right thoracic cavity (Fig. 2). The movable nodule was suspected of being thoracolithiasis but we could not confirm that it was the same nodule or that it had moved from the previous site near the esophagus. The nodule did not contain calcification, and we decided to excise the nodule for diagnostic purposes to prevent metastasis and rheumatism-related disease. Surgery was performed by thoracoscopy. A yellowish-white nodule with a diameter of 1.5 cm was found as a free body in the thoracic cavity (Fig. 3). It was easily removed from the thoracic cavity. The nodule was smooth on the surface, glossy, and the split face was yellowish-white and solid. Histopathologically, concentrically arranged collagen fibers were present, with fibroblast-like spindle-shaped cells mixed in mainly in the peripheral portion (Fig. 4). These cells were pancytokeratin positive and vimentin positive, suggesting a mesothelial origin. Granular black-brown pigment was deposited in the center; iron staining was negative, suggesting that it was charcoal powder. No adipose tissue or fat necrosis was observed. In addition, vitrification was observed, but calcification was not. In addition to this nodule, many white changes suspected of being pleural plaques were observed in the visceral pleura of the thoracic cavity. The postoperative course was uneventful, and the patient was discharged on the third postoperative day.

DISCUSSION

Thoracolithiasis is a very rare disease; the first case was reported by Dias *et al*. in 1968 [1]. Thoracolithiasis is a benign condition in which one or more free bodies with or without calcification exist in the pleural cavity without any previous history of trauma, intervention, or pleurisy [2]. Kinoshita *et al*. estimated a prevalence of 0.086% after reviewing 12,835 individuals who had undergone at least two CT scans [3]. In most cases,

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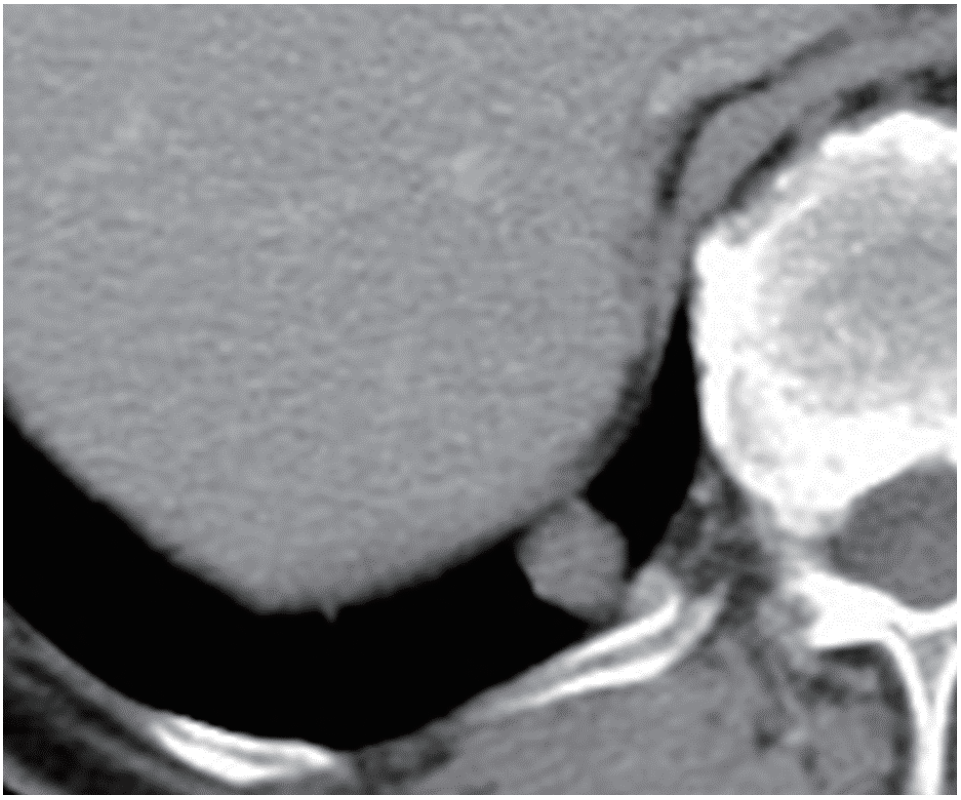

Fig. 1 Preoperative chest computed tomography (mediastinal window)

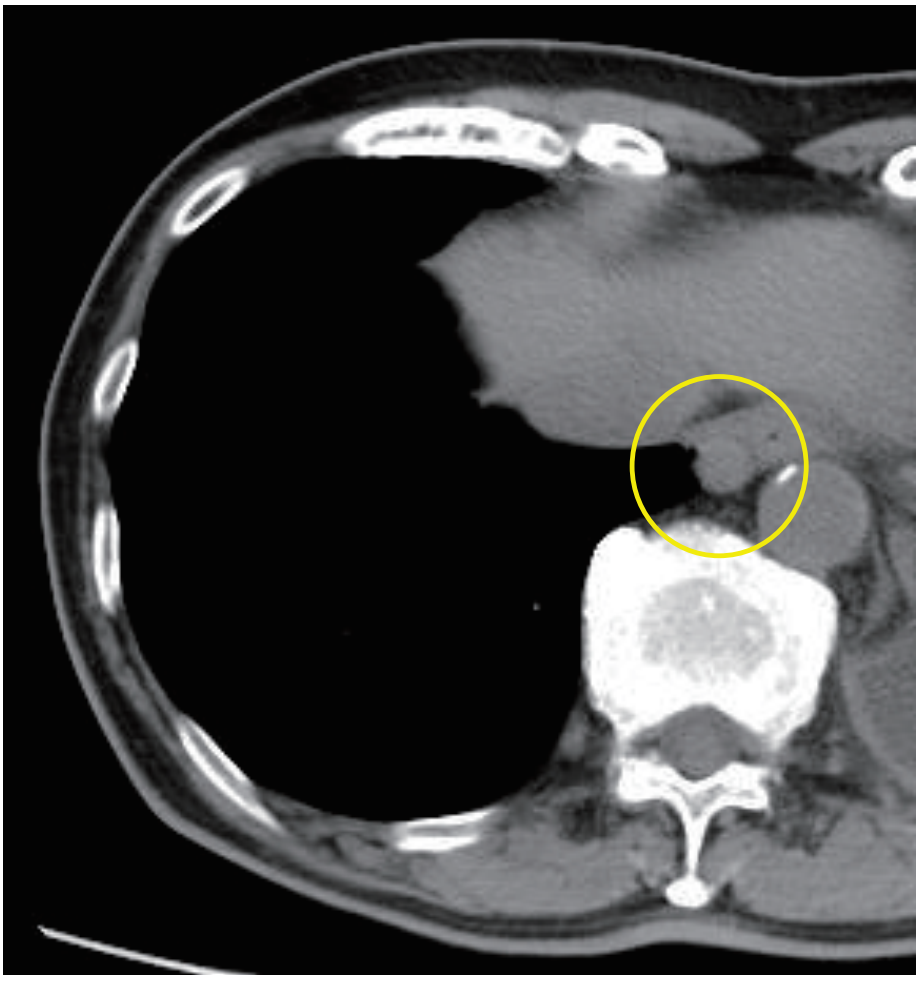

Fig. 2 Chest computed tomography 3 years before surgery (mediastinal window)

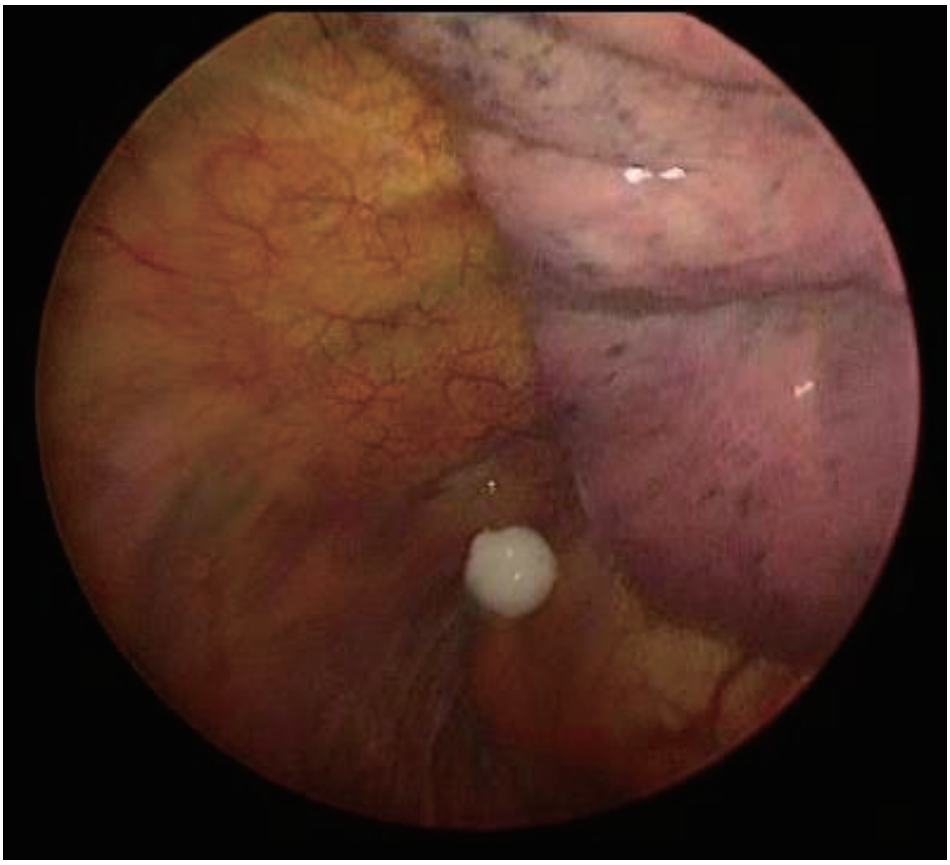

Fig. 3 Intrathoracic findings during surgery

Fig. 4 Pathological findings. Eosinophilic spherical structures with concentric circles can be seen.

thoracolithiasis is asymptomatic, and it is incidentally identified on X-ray, CT, or at the time of surgery for other diseases. The present case was asymptomatic and was identified by chance on CT during follow-up for other diseases.

Mobility on sequential imaging is the most characteristic finding of thoracolithiasis. If movement within the thoracic cavity is identified, the possibility of intrathoracic calculi is suspected before surgery. In cases with no movement, preoperative diagnostic imaging is insufficient to suspect this disease, and surgical resection is often performed for a suspected pulmonary nodule [3, 4].

There is no age or sex predilection for thoracolithiasis [4]. Seventy-five percent of cases of thoracolithiasis are described on the left side, which is explained by the fact that the left side of the thoracic cavity contains more pericardial fat than the right side [5, 6]. Thoracolithiasis is most often found in the dependent part of the pleural cavity, presumably as a result of the effect of gravity, and especially on the surface of the diaphragm, on the chest wall adjacent to the lower lung, abutting the left cardiac margin, or near to the paraspinal space. The size usually ranges from 0.5 to 1.5 cm, but the largest reported mass was 5.0 cm in diameter [1, 3]. Some cases show multiple thoracolithiasis bodies [6, 7], and some cases show a tendency for nodular size to increase [1, 2].

The exact etiology of thoracolithiasis is unclear. However, one etiological possibility is that when nuclei exist in the thoracic cavity, thickening of the fibrous capsules around these nuclei occurs, causing them to take on a tumor-like shape. The origins of these nuclei are reported as old tuberculous foci, fat tissue in the lung periphery, connective tissue formed by macrophages incorporating silica or carbon powder in the lungs, or pericardial fat necrosis tearing off in the pleural cavity [2, 5, 6, 8]. The nodule in the present case formed around carbon powder.

Plaques were also found in the visceral pleura. Some reports suggest that such plaques can tear off in the pleural cavity, forming thoracolithiasis [2]. In this case, a plaque was found in the visceral pleura, but there was no history of tuberculosis and no history of asbestos exposure.

There was no calcification in the nodule, and the patient had a history of malignant disease and rheumatoid arthritis. Metastatic lung tumor and rheumatoid nodule were also suspected. There was no reduction or movement of the nodule on CT at the follow-up 6 months after its discovery. We were unable to confirm that the nodule in the ipsilateral thoracic cavity identified 3 years prior was the same nodule as the present nodule. The nodule was relatively immobile because it was in the lower part of the thoracic cavity.

In conclusion, the pathological significance of thoracolithiasis is not clear. Surgery should be considered for cases where it is difficult to distinguish it from other diseases or in cases where nodular size increases.

DISCLOSURE

The authors have no conflicts of interest to declare.

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AUTHOR CONTRIBUTIONS

All the authors approved the manuscript.

ETHICS STATEMENT

This case report is for academic communication only and not for other purposes. The patient provided informed consent, and patient anonymity was preserved. The approval of our imstitutional ethics committee was unnecessary for a clinical case report.

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