

Surgically Removed Thoracolithiasis: Report of Two Cases

Teruo Iwasaki, MD, PhD,¹ Katsuhiko Nakagawa, MD, PhD,¹ Hiroshi Katsura, MD,¹
Naoko Ohse, MD,¹ Teruaki Nagano, MD,² and Kunimitsu Kawahara, MD, PhD²

Thoracolithiasis is a rare condition with only 12 cases of surgically removed nodules reported in the literature. We report 2 additional cases.

Case 1: A 19-year-old male admitted with an abnormal shadow on a chest X-ray. Computed tomography (CT) revealed a nodule in the right lower lung lobe. The material extirpated by thoracoscopy was milky white, glossy, and 1.6 cm in diameter. Histopathologically, it consisted of fatty necrotic tissue covered with hyalinized fibrous tissue.

Case 2: A 78-year-old female, with a past history of breast cancer, admitted with an abnormal shadow on chest X-ray. CT revealed a nodule in the left lung S¹⁺² segment, of which transbronchial biopsy findings indicated primary lung adenocarcinoma. Exploratory thoracoscopy incidentally revealed some pearly material, 0.4 cm in diameter, in the thoracic cavity. They were extirpated during left upper lobectomy for lung cancer; all of them demonstrated concentric hyalinized fibrous tissue. Thoracic surgeons should consider this condition in the differential diagnosis of a peripheral pulmonary nodule. (*Ann Thorac Cardiovasc Surg* 2006; 12: 279–82)

Key words: thoracolithiasis, pleural stone, thoracoscopy

Introduction

Thoracolithiasis, which is also described as pleural stone, intrathoracic calculus or pleurolith, is a rare condition and only 12 cases of surgically removed nodules have been reported in the literature.^{1–11)} We report 2 additional cases of surgically removed thoracolithiasis. Thoracic surgeons should bear in mind this condition in the differential diagnosis of a peripheral pulmonary nodule.

Cases

Case 1

A 19-year-old otherwise healthy male was admitted for

From Departments of¹Respiratory Surgery and²Pathology, Osaka Prefectural Medical Center for Respiratory and Allergic Diseases, Habikino, Japan

Received October 28, 2005; accepted for publication December 24, 2005.

Address reprint requests to Teruo Iwasaki, MD, PhD: Department of Respiratory Surgery, Osaka Prefectural Medical Center for Respiratory and Allergic Diseases, 3–7–1 Habikino, Habikino, Osaka 583–8588, Japan.

further evaluation of an abnormal shadow found on chest X-ray. On admission, physical examination and laboratory data were unremarkable. A chest X-ray showed a well-defined oval nodule 1.5 cm in diameter in the right lower lung field (Fig. 1A). Computed tomography (CT) of the chest revealed a subpleural lesion, within which minute high density areas were detected in the lung window (Fig. 1B), but the nodule disappeared in the mediastinal window (Fig. 1C). A benign lung tumor such as a hamartoma was suspected.

Video-assisted thoracic surgery (VATS) was performed to examine the surface of S⁹ segment of the lung with a scope from the 7th intercostal space (ICS) on the midaxillary line and to remove the material with a forceps from the 6th ICS on the posterior axillary line. The extirpated material was 1.6×1.5 cm in size and its surface was milky white and glossy (Fig. 2A); its cross-section was rough and yellowish brown (Fig. 2B). Histopathologically, it consisted of adipose and fatty necrotic tissue surrounded by hyalinized fibrous tissue (Figs. 2C and 2D). The patient's postoperative course was uneventful.

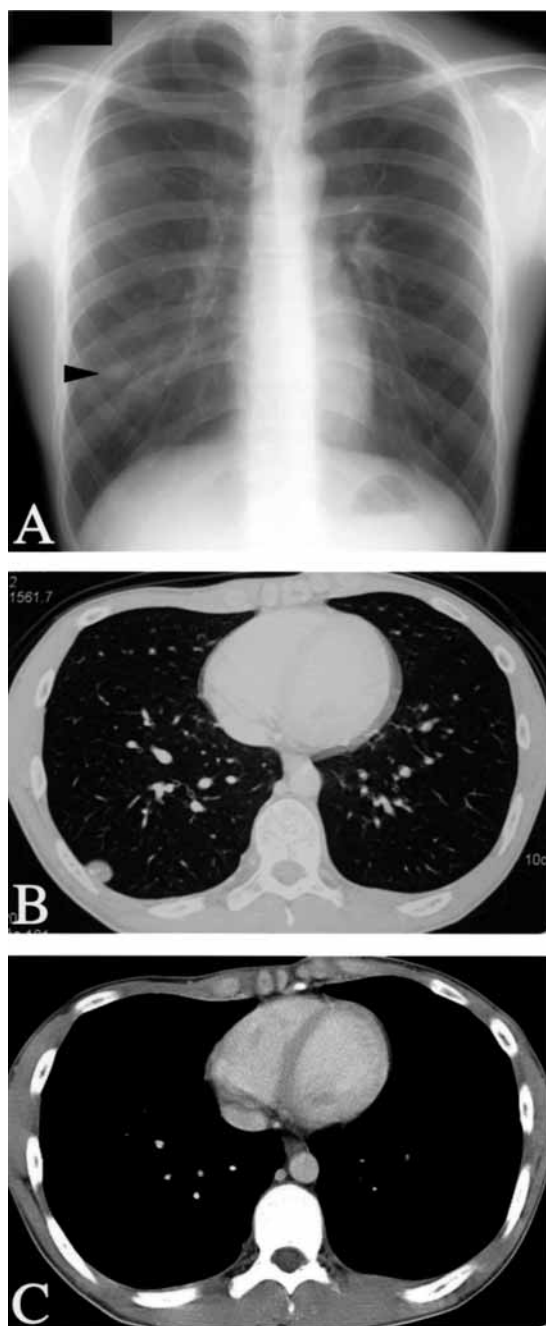


Fig. 1.

- A:** Chest X-ray showing a well-defined oval nodule (arrowhead) in the right lower lung field.
- B:** Minute high density areas were detected within the nodule in the lung window of chest CT.
- C:** The nodule disappeared in the mediastinal window.

Case 2

A 78-year-old female was admitted to our hospital for further evaluation of an abnormal shadow detected by chest screening X-ray. She had undergone a left radical

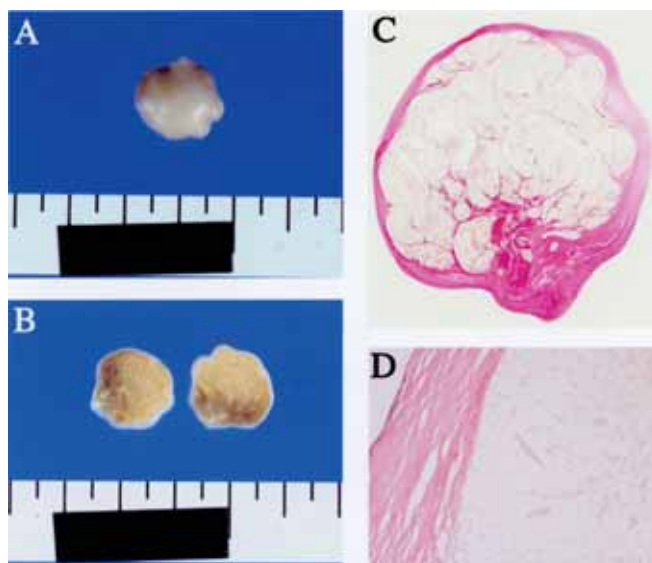


Fig. 2.

- A:** Photograph of the extirpated material. It was 1.6×1.5 cm in size and its surface was milky white and glossy.
- B:** The cross-section of the material was rough and yellowish brown, and the contents were covered with a milky white capsule.
- C, D:** Microscopic findings of the extirpated material under low (C; ×4.7) and high (D; ×40) power magnifications (HE stain) in case 1. The material consisted of adipose and fatty necrotic tissue surrounded by hyalinized fibrous tissue.

mastectomy for breast scirrhous carcinoma (pT2N3cM0 stage IIIc, estrogen- and progesterone-receptor negative) at the age of 70, followed by chemotherapy (cyclophosphamide plus methotrexate plus fluorouracil and doxorubicin plus medroxyprogesterone acetate) and radiotherapy (50 Gy) over the left supraclavicular and parasternal regions. A chest X-ray and CT demonstrated a nodule in the left lung S¹⁺² segment, and transbronchial biopsy findings of the lesion indicated primary lung adenocarcinoma.

An exploratory thoracoscopy was performed through the 7th ICS on the midaxillary line to incidentally find some pearly material on funicular and membranous adhesions between the parietal and visceral pleurae without disseminated lesions (Fig. 3). A left posterolateral incision and thoracotomy through the 4th ICS were done to carry out a complete left upper lobectomy for lung cancer. Histopathological examination revealed the lung cancer to be moderately differentiated adenocarcinoma (pT2N0M0 stage IB). Some pearly materials were extirpated and all of them were found to be concentric hyalinized fibrous tissue (Fig. 4). The postoperative course was uneventful.

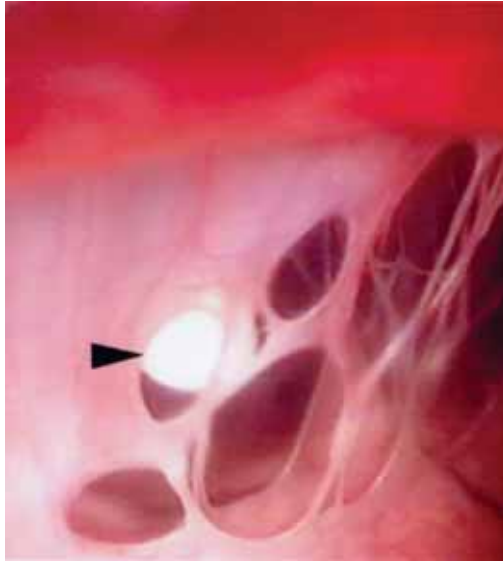


Fig. 3. Exploratory thoracoscopy revealed the presence of pearly material (arrowhead) on funicular and membranous adhesions between the parietal (top) and visceral (bottom) pleurae.



Fig. 4. Microscopic findings of one of the extirpated materials, 0.4 cm in diameter, in case 2. The material consisted of concentric hyalinized fibrous tissue. (HE stain: ×20)

Table 1. Reports of surgically removed thoracolithiasis in Japan

Case	Author/ref. no.	Year	Age (years)	Gender	Site	Preoperative detection	Mobility	Calcification	Size (cm)
1	Takiguchi et al. ²⁾	1987	71	M	L	+	+	+	1.5×1.2×0.8
2	Ishikawa et al. ³⁾	1988	57	M	L	-	nd	+	1×1×0.4
3	Kuwabara et al. ⁴⁾	1989	56	M	L	-	nd	-	0.6
4	Fujiwara et al. ⁵⁾	1992	53	F	R	+	+	-	1.5×1.2×0.7
5	Kuroya et al. ⁶⁾	1996	50	F	L	+	-	-	2×1×1
6	Kosaka et al. ⁷⁾	2000	76	M	L	+	-	-	1.5
7	Kosaka et al. ⁷⁾	2000	54	F	L	-	nd	-	0.5
8	Ando et al. ⁸⁾	2002	67	M	L	+	-	-	1.7×1.5×1.0
9	Takeichi et al. ⁹⁾	2004	72	M	L	+	-	+	2.5×2.0×1.8
10	Ito et al. ¹⁰⁾	2005	80	M	R	+	+	-	2.2×1.8
11	Our case	2006	19	M	R	+	-	-	1.6×1.5
12	Our case	2006	78	F	L	-	nd	-	0.4

M, male; F, female; L, left; R, right; nd, not determined.

Discussion

Dias et al.¹⁾ reported the first case of pleural stone in 1968 and noted that no similar cases had been found in the literature until then. Takiguchi et al.²⁾ termed an unusual isolated calcified lesion in the intrathoracic space “thoracolithiasis”, and Kosaka et al.⁷⁾ defined thoracolithiasis as a condition in which 1 or more free bodies with or without calcification exist in the thoracic cavity without any previous trauma, intervention, or pleurisy. To the best of our knowledge, only 12 cases of surgically removed nod-

ules have been reported in the literature: 10 in Japan,²⁻¹⁰⁾ 1 in America¹⁾ and 1 in Germany.¹⁰⁾ In 3 cases,^{3,9,11)} the patients had a previous history of pleurisy and 1 of them had also undergone an artificial pneumothorax for pulmonary tuberculosis.³⁾ Here we consider the above-mentioned condition as thoracolithiasis regardless of pleurisy.

The characteristics of the 12 Japanese cases, including our 2 cases, are summarized in Table 1. Two thirds were men (67%; 8 cases). The patients' age ranged from 19 years to 80 years (mean: 61 years): our case 1 was the youngest. Nine cases (75%) occurred in the left hemitho-

rax. The maximum diameter of the material ranged from 0.4 cm to 2.5 cm (median: 1.5 cm). All patients were asymptomatic except for 1 case in which the material shadow gradually enlarged and the patient complained of productive cough.⁷⁾

Four cases, including our case 2, had no imaging evidence before extirpation, the nodules were discovered incidentally during surgery for lung cancer, and were no more than 1 cm in diameter (median: 0.6 cm).^{3,4,7)} Eight cases were detected on chest X-ray and/or CT before extirpation and were at least 1.5 cm in diameter (median: 1.6 cm). In 3 of these 8 cases, thoracolithiasis was noted to be mobile during the course of their evaluation.^{2,5,10)} Five cases, including our case 1, were immobile and had been preoperatively diagnosed as a peripheral pulmonary tumor. These findings showed that thoracolithiasis was difficult to diagnose correctly when they were small (<1 cm) or immobile. Thoracoscopy was reported to be useful for the diagnosis and treatment of thoracolithiasis, and the same was true in our cases.

Histopathological findings of the extirpated materials were as follows: fibrous tissue with fatty necrosis at the core in 7 cases (including our case 1); calcification covered with fibrous tissue in 1 case;⁹⁾ fatty tissue with calcification in 1 case;²⁾ fibrous tissue with caseous necrosis at the core in 1 case;³⁾ fibrous tissue with dust, containing calcium compounds, at the core in 1 case;⁶⁾ and hyalinized fibrous tissue in 1 case (our case 2). Thus, thoracolithiasis usually consisted of fatty tissue with or without necrosis (8 cases; 67%) and/or calcification or calcium compounds (4 cases; 33%). Magnetic resonance imaging (MRI) of the chest was done only in 1 surgical case.¹⁰⁾ Both T1- and T2-weighted MRI revealed a central area of high intensity corresponding to fatty necrotic tissue. The histological characteristics of the thoracolithiasis described above suggest the diagnostic usefulness of MRI.

The etiology remains to be clarified. However, some explanations for the core formation in thoracolithiasis have been proposed: (1) pleural or pericardial fat dropping into the intrathoracic space;⁷⁻¹⁰⁾ (2) pleural or peripheral pulmonary lipoma tearing off;^{2,4)} (3) focus of old pulmonary tuberculosis;^{3,9)} and (4) aggregation of macrophages phagocytosing dust.⁶⁾ The relationship between pericardial fat and thoracolithiasis is supported by a predominant (75%) left hemithorax occurrence. Inflammation may also facilitate the fibrosis and development of thoracolithiasis, as in our case 2. Its association with chemotherapy, radiotherapy or concomitant lung cancer is unknown. The elucidation of its etiology requires the accumulation of

additional cases.

Conclusion

We report 2 additional cases of surgically removed thoracolithiasis. This condition is difficult to diagnose correctly in immobile cases even if detectable. Since it usually consists of fatty tissue or calcification at the core, MRI is useful for its diagnosis, in addition to thoracoscopy. Thoracic surgeons should consider this condition in the differential diagnosis of a peripheral pulmonary nodule.

References

1. Dias AR, Zerbini EJ, Curi N. Pleural stone. A case report. *J Thorac Cardiovasc Surg* 1968; **56**: 120-2.
2. Takiguchi Y, Hashizume I, Shinozaki K, Yasuda J, Hanzawa S, Kadoyama C. A case of "thoracolithiasis"—an unusual isolated calcified lesion in the intrathoracic space. *Nihon Kyobu Shikkan Gakkai Zasshi* 1987; **25**: 776-80. (in Jpse.)
3. Ishikawa S, Genna K, Kawabata T, Maesato K, Kuniyoshi M, Yamauchi K. A case of "thoracolithiasis". *J Nat Oki Hos* 1988; **9**: 33-5. (in Jpse.)
4. Kuwabara M, Okumura N, Fukuse T, Kou T, Ariyasu T. A case showing a free substance in the thoracic cavity detected during surgery in lung cancer considered to the precursor of the so-called "intrathoracic calculus". *Nihon Kyobu Shikkan Gakkai Zasshi* 1989; **27**: 730-4. (in Jpse.)
5. Fujiwara A, Akaogi E, Yamamoto T, et al. A case of "migrating thoracolithiasis". *Jpn J Chest Dis* 1992; **51**: 1063-6. (in Jpse.)
6. Kuroya M, Inui K, Yokomise H, et al. Floating object in pleural space, a precursor of thoracolithiasis; a case report. *J Jpn Assoc Chest Surg* 1996; **10**: 52-6. (in Jpse.)
7. Kosaka S, Kondo N, Sakaguchi H, Kitano T, Harada T, Nakayama K. Thoracolithiasis. *Jpn J Thorac Cardiovasc Surg* 2000; **48**: 318-21.
8. Ando K, Shimamura T, Kurisu S, Yamamura T, Takakuwa T, Osada H. A case of thoracoscopically removed intrathoracic stone. *J Jpn Surg Assoc* 2002; **63**: 1122-5. (in Jpse.)
9. Takeichi H, Masuda R, Yoshino K, et al. A case of thoracolithiasis. *J Jpn Assoc Chest Surg* 2004; **18**: 759-63. (in Jpse.)
10. Ito Y, Koike Y, Yoneyama S, Kon H, Sasaki A. A case of thoracolithiasis extirpated. *J Jpn Surg Assoc* 2005; **66**: 618-21. (in Jpse.)
11. Schneider V, Zimmermann HD, Walz L. Über Corpora libera der Pleurahöhle. *Röfo* 1970; **113**: 437-42. (in German)