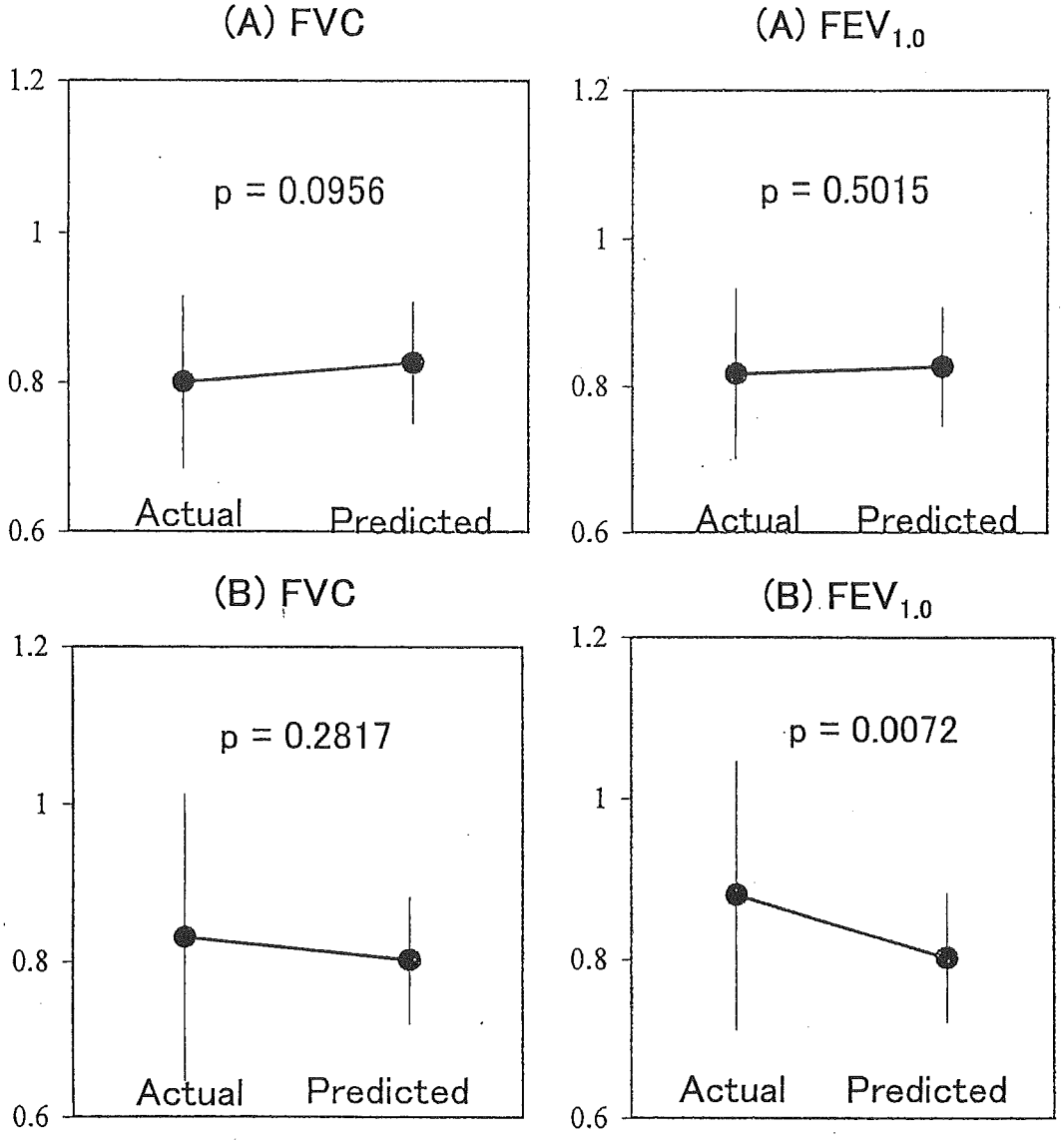

Figure 1. Student's *t*-test result of predicted and actual postoperative residual rate of FVC and $FEV_{1.0}$ in patients without pulmonary parenchymal disease and with emphysema. (**A**): no respiratory disease, (**B**): emphysema

において術後実測残存率は術後予測残存率より有意に高値を示した．さらに，E群で血流シンチグラム上，切除部位が血流の低下部位と一致した症例においては，術後実測残存率はFVC，$FEV_{1.0}$ともに術後予測残存率より高値を示した．また今回の対象の104例には，画像所見上，腫瘍により末梢側が無気肺を呈した症例は存在せず，肺血流シンチグラムの血流低下部位は気腫性変化によるものと考えられた．以上より肺気腫合併症例において，気腫性変化の強い部位が肺癌切除部位と同一の症例では，LVRS効果が得られ，予測よりも術後の残存肺機能が保たれる可能性が示唆された．

先に述べたように肺癌の手術適応として術後予測$FEV_{1.0}$ 800 ml以上という基準が提唱されている[7-9]が，今回の検討例で2例が手術後の予測$FEV_{1.0}$が800 ml以下であった．いずれも肺気腫合併例であったが，血流低下部位と肺癌の存在部位が一致するために手術を施行し，完全切除（肺葉切除術1例，区域切除術1例）が可能となった．1例は術前の$FEV_{1.0}$ 1.08 l，1秒率40.1%で右下葉切除術を施行した．術後の予測$FEV_{1.0}$が778 mlであったが，実測値は$FEV_{1.0}$ 1.03 l，1秒率68.0%と術後予測値より改善した．もう1例は術前の$FEV_{1.0}$ 1.01 l，1秒率41.0%で右底区域切除術を施行した．術後の予測$FEV_{1.0}$が788 mlであったが，術後の実測値は$FEV_{1.0}$ 1.01 l，1秒率51.8%と，術前の予測より高値を示した．

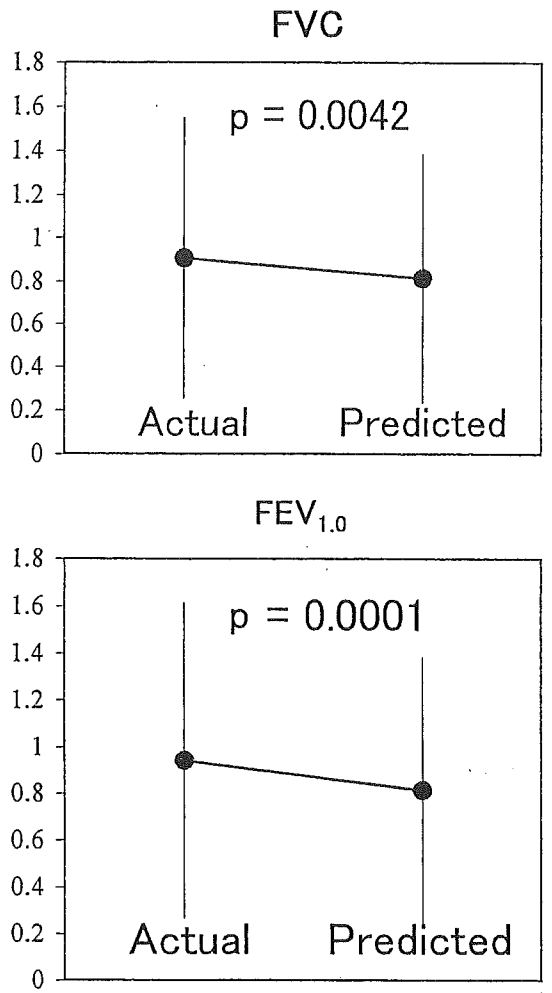

Figure 2. Student's *t*-test result of predicted and actual postoperative residual rate of FVC and $FEV_{1.0}$ in 23 patients with emphysema whose resection areas matched their target areas.

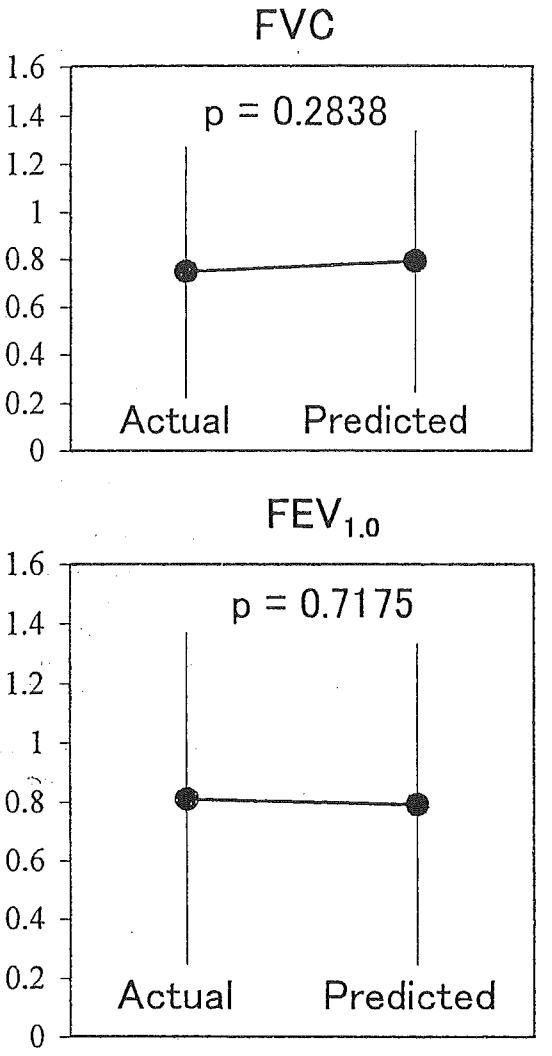

Figure 3. Student's *t*-test result of predicted and actual postoperative residual rate of FVC and $FEV_{1.0}$ in 20 patients with emphysema whose resection areas did not match their target areas.

この2例はいずれも血流シンチグラム上，血流分布が不均一で，腫瘍の存在部位が血流の低下部位と一致しており，またCT上も腫瘍の存在する下葉の気腫性変化が強かった．このため手術後の予測 $FEV_{1.0}$ が 800 ml 以下であったが，LVRS 効果が十分得られると考え，また病期からも手術療法が一番根治につながると判断した．幸い，術後合併症もなく，安全に完全切除術が施行できた．

肺気腫合併肺癌症例は，たとえ低肺機能であっても切除部位が血流低下部位と一致するならば，LVRS 効果により予測残存肺機能より実際の残存肺機能が保たれることが期待でき，標準手術や切除術の対象となりうると考えられた．今後は術前肺機能，血流シンチグラムや Thin-slice-CT を含む画像所見から肺気腫の存在を予測し，肺気腫の程度により術後肺機能を予測できれば，臨床に適応できると考えられる．

結　語

肺気腫合併肺癌症例では術後残存 $FEV_{1.0}$ が予測値よりも高くなる場合があり，特に切除部位に血流低下を認める症例では LVRS 効果が得られると推察された．肺気腫合併低肺機能例でも LVRS 効果を期待し，手術適応を検討すべきと考える．

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DOI 10.1007/s00428-005-0074-4

LETTER TO THE EDITOR

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Katsuhiro Nakagawa · Hiroshi Katsura ·
Teruaki Nagano · Teruo Iwasaki

Multiple calcifying fibrous tumor of the pleura

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Dear Sir,

The article by Mito et al. [4] concerning multiple calcifying fibrous tumor of the pleura (CFTP) is a great contribution to the subject of respiratory pathology, as only eight cases of CFTP have been reported [2–4], and multiple CFTP has been poorly documented. In our opinion, pathologists and surgeons should be made aware of multiple CFTP since it is possible to confuse CFTP macroscopically with disseminated metastatic sarcoma or diffuse malignant mesothelioma during surgery. We would like to add one additional case of multiple CFTP that was pathologically diagnosed on intraoperative frozen section slides.

Case report

A 35-year-old Japanese woman, who had smoked about 20 cigarettes per day for 15 years, consulted our hospital complaining of right chest pain that had occurred for the past 2 years. Occupational exposure to asbestos was probable. Computed tomographic scan of the chest showed remarkable thickening of the parietal pleurae of the right hemithorax. A tentative diagnosis of diffuse malignant mesothelioma was made. Family history and past medical history were unremarkable. Video-assisted thoracoscopic surgery was performed for histological examination. Thoracoscopic examination demonstrated a large arborized nodule, 5 cm in diameter on the diaphragmatic pleura (Fig. 1a), and other miliary dome-shaped nodules at the costal and mediastinal pleurae as well as visceral pleura mainly in the lower lobe (Fig. 1b). There was a small amount of serous pleural effusion. In the pleural fluid, no cytological abnormalities were observed. Pleural plaques were present at the fourth, fifth, and sixth costal pleura. The surgeons thoracoscopically diagnosed eccentric disseminated metastatic sarcoma or diffuse malignant mesothelioma at resection of the largest nodule. Intraoperative frozen section slide showed hyalinized fibrosclerotic nodular lesions with microcalcifications, which highly supported the pathological diagnosis of multiple CFTP. Thoracotomy was performed, and all remaining nodules were extirpated. On postoperative gross examination, all nodules were well circumscribed, solid, or firm with a uniform gray-white fibrous cut appearance without any hemorrhage or necrosis (Fig. 1c). All nodules were pleural-based. Histologically, the nodules were circumscribed, nonencapsulated, and composed chiefly of hypocellular hyalinized fibrosclerotic tissue with scattered cytologically bland spindle cells, scant lymphoplasmacytic infiltrate, and small calcifications. The calcifications had a laminated appearance showing typical psammoma bodies (Fig. 2a). The surface of the nodules was occasionally covered by a single layer of mesothelial cells. Asbestos bodies were not detected in or around the nodules. Immunohistochemical studies showed that most spindle cells were positive for vimentin and factor XIIIa (Fig. 2b) but negative for epithelial membrane antigen (EMA), keratin (AE1/3), desmin, α-smooth muscle actin, S-100, anaplastic lymphoma kinase, and CD34. Taken together, a pathological diagnosis of multiple CFTP was made.

Recognition of this extremely rare miliary type of CFTP is important because of the possibility of confusing it intraoperatively with malignant fibrous neoplasms including desmoplastic mesothelioma or disseminated metastatic sarcoma [2–4]. Desmoplastic mesothelioma shows a storiform collagen pattern, collagen necrosis, bland acellular collagen, and focal cytologic features of malignancy [5]. Moreover, desmoplastic mesothelioma stains for cytokeratins, but our case was negative for AE1/3. In disseminated metastatic fibrous sarcoma with pleural

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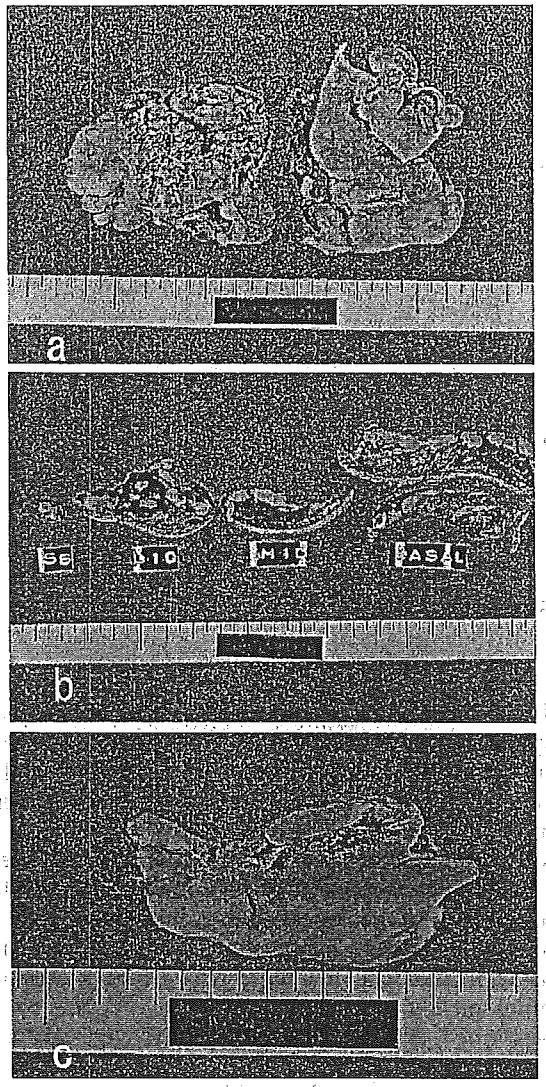

Fig. 1 a A large arborized nodule measuring 5 cm in diameter was present on the diaphragmatic pleura. b Miliary dome-shaped nodules were also present at the visceral pleura of the right lower lobe. c The cut surface of the largest nodule showed a uniform fibrous appearance

calcification, monophasic synovial sarcoma (MSS) should be considered. MSS is usually a localized pleural-based mass [1]. MSS is composed of nearly uniform spindle fibroblast-like cells that are arranged in dense cellular sheets or vague fascicles. Hyalinization and calcification are occasionally seen. In our case, dense cellular areas composed of fibroblast-like cells were absent. Immunohistochemically, cytokeratin and/or EMA is positive in MSS, but this finding was negative in our patient.

In this case, we made a diagnosis of multiple CFTP based on intraoperative frozen section slides. The keys to accurate diagnosis were as follows: (1) recognition of submesothelial origin; (2) hypocellular hyalinized fibrosclerotic tumor with psammoma bodies; and (3) knowledge about multiple CFTP obtained from the article by Mito et al. [4].

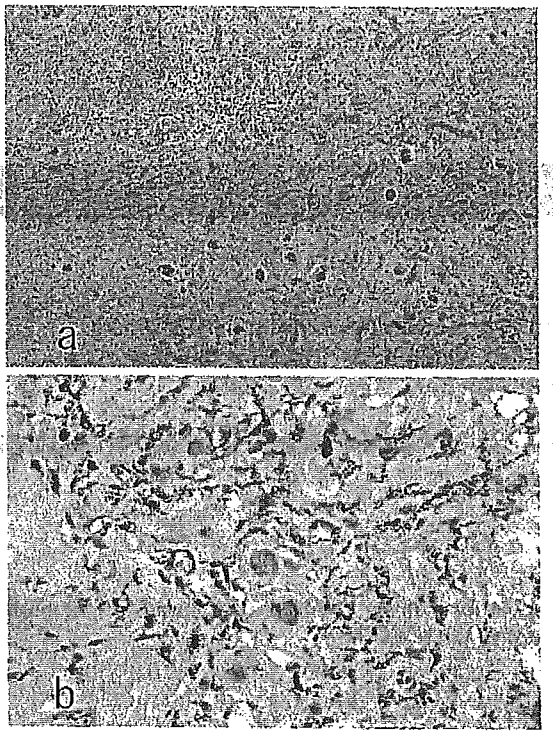

Fig. 2 a Histologically, the tumor was predominantly composed of hypocellular hyalinized fibrosclerotic tissue with psammoma bodies and scant lymphoplasmacytic infiltrate (×50). b The spindle cells showed positive staining for factor XIIIa immunohistochemically (×100)

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Surgically Removed Thoracolithiasis –Report of Two Cases

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Key words: thoracolithiasis, pleural stone, thoracoscopy

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Comments for transmission to the authors should be typed on this sheet.

Authors: Teruo Iwasaki ,et al.

REFEREE'S RECOMMENDATION

[V] Accept
[ ] Accept with minor changes
[ ] Accept with revision as per attached comments
[ ] Reject

REFEREE'S COMMENTS

(Japanese comments are acceptable only when authours are Japanese)

This paper is now accepted, because all questions were clarified.

However, Figures should be more summarized as follows.

Fig. 3 A.B → Fig. 2 C D, because of demonstration of case 1

Fig. 4 → Fig. 3

Fig. 5 → Fig. 4

Ann. Thorac. Cardiovasc. Surg.

Two Cases of Thoracolithiasis Extirapated

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Key words: thoracolithiasis, pleural stone, thoracoscopy

Abstract

Thoracolithiasis is a rare condition with only 12 extirpated cases reported in the literature. We here report 2 additional cases. Case 1: A 19-year-old man was admitted with an abnormal shadow on a chest X-ray. A computed tomography (CT) demonstrated a nodule in the right lower lobe. The material extirpated by thoracoscopy was milky white, glossy, and 1.6 cm in diameter. Microscopic examination showed it consisting of fatty necrotic tissue covered with hyalinized fibrous tissue. Case 2: A 78-year-old woman, with past history of breast cancer, was admitted with an abnormal shadow on a chest X-ray. CT demonstrated a nodule in the left S^{1+2} segment, of which transbronchial biopsy revealed primary lung adenocarcinoma. Exploratory thoracoscopy incidentally revealed some pearly materials, 0.4 cm in diameter, in the thoracic cavity. They were extirpated during left upper lobectomy for lung cancer, and found all concentric hyalinized fibrous tissue.

Introduction

Thoracolithiasis, also described as pleural stone, intrathoracic calculus or pleurolith, is a rare condition and only 12 extirpated cases have been reported in the literature.[1–10] We here report 2 additional cases of thoracolithiasis extirpated and, in a detectable case, would like to remind thoracic surgeons of this condition in the differential diagnosis of a peripheral pulmonary nodule.

Cases

Case 1. A 19-year-old otherwise healthy man was admitted for further evaluation of an abnormal shadow found on a 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). A computed tomographic (CT) scan of the chest demonstrated a subpleural lesion, within which minute high density areas were detected in the lung window (Fig. 1B), and 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 observe an impression on the surface of S^9 segment of the lung with a scope from the 7th intercostal space (ICS) on the midaxillary line and to remove the material in the paravertebral lesion with a forceps from the 6th ICS on the posterior axillary line. The extirpated material was milky white and glossy, with size of 1.6 × 1.5 cm (Fig. 2A), and its cross-section was rough and yellowish brown (Fig. 2B). Microscopic examination showed that it consisted of adipose and fatty necrotic tissue surrounded by hyalinized fibrous tissue (Fig. 3A and B). The patient's postoperative course was uneventful.

Case 2. A 78-year-old woman was admitted to our hospital with an abnormal shadow in a chest screening X-ray. She had undergone a left radical mastectomy for breast scirrhous carcinoma (pT2N3cM0 Stage IIIc, estrogen- and progesterone-receptor negative) when 70 years old, followed by chemotherapy (cyclophosphamide plus methotrexate plus fluorouracil and doxifluridine plus medroxyprogesterone acetate) and radiotherapy (50 Gy) to the left supraclavicular and parasternal regions. A chest X-ray and CT demonstrated a nodule in the left S^{1+2} segment, and transbronchial biopsy of the lesion revealed primary lung adenocarcinoma.

Exploratory thoracoscopy was performed through the 7th ICS on the midaxillary line to find incidentally some pearly materials on funicular and membranous adhesions between parietal and visceral pleurae (Fig. 4) without disseminated lesions. A left posterolateral incision and thoracotomy through the 4th ICS was done to complete left upper lobectomy for lung cancer. Pathological examination revealed the lung cancer to be moderately differentiated adenocarcinoma (pT2N0M0 Stage IB). Some pearly materials were extirpated and found all concentric hyalinized fibrous tissue (Fig. 5). The patient ran an uneventful postoperative course.

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 one 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 extirpated cases have

been reported in the literature: 10 were in Japan,[2–9] 1 in America[1] and 1 in Germany.[10] In 3 cases,[3, 8, 10] the patients had the previous history of pleurisy and 1 of them also underwent an artificial pneumothorax for pulmonary tuberculosis.[3] Here we consider the above-mentioned condition as thoracolithiasis regardless of pleurisy.

The characteristics of 12 Japanese cases, including our 2, are summarized as follows. They are more common in men (67%; 8 cases) than in women (33%; 4 cases). Patients' ages ranged from 19 to 80 years (mean: 61 years): our patient in Case 1 was the youngest. Nine cases (75%) occurred in the left hemithorax. 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 sputum.[7]

Four cases, including Case 2, had no imaging evidence before extirpation, 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.[2, 5, 9] Five cases,

including Case 1, were immobile and preoperatively diagnosed as a peripheral pulmonary tumor. These findings show that thoracolithiasis is difficult to diagnose correctly in smaller (<1 cm) or immobile cases. Thoracoscopy was reported to be useful in diagnosis and treatment of thoracolithisis, and the same was true of our cases.

Histological findings of the extirpated materials were as follows: fibrous tissue with fatty necrosis at the core in 7 cases (including Case 1); calcification covered with fibrous tissue in 1 case;[8] fatty tissue with calcificasion in 1 case;[2] fibrous tissue with caseous necrosis at the core in 1 case;[3] fibrous tissue with dust, containig calcium compounds, at the core in 1 case;[6] and hyalinized fibrous tissue in 1 case (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 extirpated case.[9] On both T1- and T2-weighted imaging, MRI revealed a central high intensity corresponding to fatty necrotic tissue at the core. Histological characteristics, as described above, of the thoracolithiasis suggest the diagnostic usefulness of MRI.

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;[7–9] (2) pleural or peripheral pulmonary lipoma tearing off;[2, 4] (3) focus of old pulmonary tuberculosis;[3, 8] and (4) aggregation of macrophages phagocyting dust.[6] The relationship between pericardial fat and thoracolithiasis will be supported by a predilection (75%) for the left hemithorax, although Case 1 occurred in the right. Inflammation such as pleuritis may also facilitate the fibrosis and development of thoracolithiasis, as in 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 here report 2 additional cases of thoracolithiasis extirpated. Thoracolithiasis is difficult to diagnose correctly in immobile cases even if detectable. Since it usually consists of fatty tissue or calcification at the core, MRI will be of great use in its diagnosis besides thoracoscopy. Thoracic surgeons have to think of this condition in the differential diagnosis of a peripheral pulmonary nodule.

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Figure legends

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.

Fig. 2 A: Photograph of the extirpated material, the surface of which was milky white and glossy, with size of 1.6 × 1.5 cm. B: The cross-section of the material was rough and yellowish brown, and showed that the contents were covered with a milky white capsule.

Fig. 3 Microscopic findings of the extirpated material under low (A; ×4.7) and high (B; ×40) power magnifications (HE staining) in Case 1. The material consisted of adipose and fatty necrotic tissue surrounded by hyalinized fibrous tissue.

Fig. 4 Thoracoscopic findings of the pearly material (arrowhead) on funicular and membranous adhesions between parietal (top) and visceral (bottom) pleurae.

Fig. 5 Microscopic findings of one of the extirpated materials, 0.4 cm in diameter, showed hyalinized fibrous tissue (×20, HE staining) in Case 2.

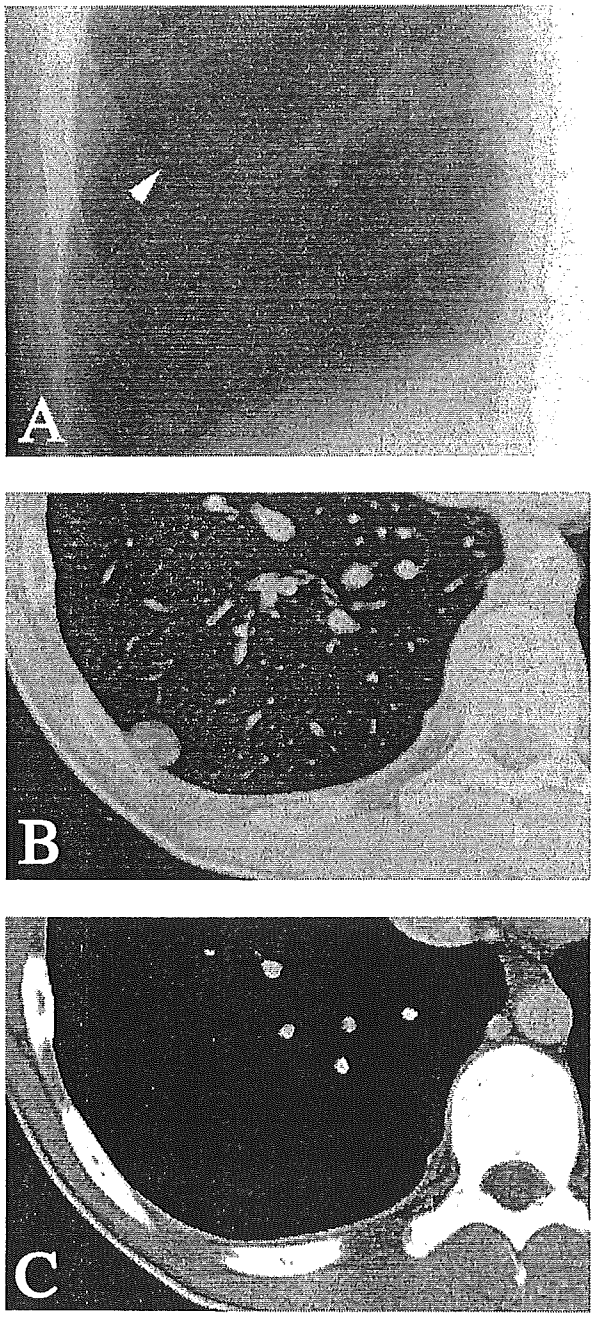

Fig. 1

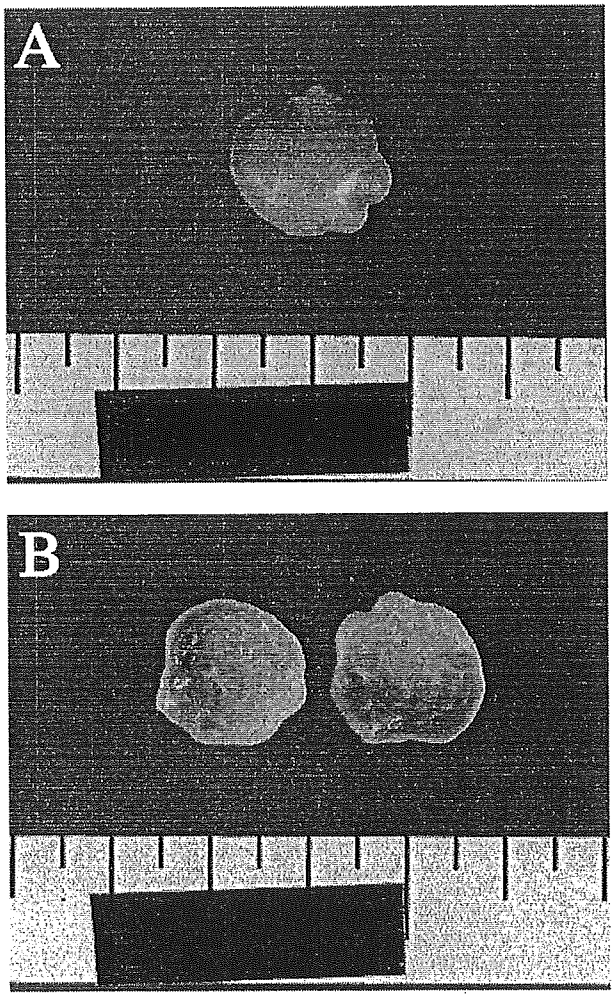

Fig. 2