



Case report

A rare case of numerous thoracolithiasis with chest discomfort

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ABSTRACT

Thoracolithiasis is a rare benign condition with mobile free bodies in the pleural cavity. It is asymptomatic and mostly found incidentally. Up to our knowledge there is no report of symptomatic numerous thoracolithiasis. We report a very rare case of thoracolithiasis in a 36-year-old female with chest discomfort. Images from computed tomography presented a chain of small non-enhancing nodules in the left hemi-diaphragmatic pleura. Exploratory thoracoscopy was performed and twenty-five mobile pearl like thoracolithiasis were discovered. Histopathology showed extensive necrotic fatty tissue at its center surrounded by fibrosis. The patient was symptom-free after the surgical removal of numerous thoracolithiasis, suggesting thoracolithiasis was associated with chest discomfort.

1. Introduction

Thoracolithiasis is a rare benign condition in which one or more mobile free bodies with or without calcification exist in the pleural cavity without any previous history of trauma, intervention or pleurisy [1]. It can manifest as one or two small nodules mostly without symptoms and are incidentally found on a chest image or during thoracic surgery. Up to our knowledge there is no report of symptomatic numerous thoracolithiasis. Here we report a very rare case of multiple thoracolithiasis in a 36-year-old female presented with chest discomfort.

2. Case report

A 36-year-old female was referred to our hospital for further evaluation on abnormal finding in the computed tomography (CT). The patient had chest discomfort and mild dyspepsia for 3 months but otherwise healthy. She never smoked, had no previous medical history including trauma or pulmonary disease including tuberculosis. Her blood laboratory tests were unremarkable. In the primary clinic, chest imaging evaluation was done. On axial image of chest CT, there was a chain of non-enhancing small nodules in the left hemi-diaphragmatic pleura with fat containing lesion (Fig. 1A and B). The chest magnetic

resonance imaging (MRI) also showed similar findings from chest CT without chest wall invasion (Fig. 1C). On positron emission tomography-computed tomography (PET-CT) represented pleural based lesions without significant FDG uptake in left lower lobe of the lung (Fig. 1D). The leading initial radiographic impression was pleural lipomatosis and was referred to thoracic surgeon to evaluate the necessity of surgical resection. Due to patient's symptom and growing rate of tumor, the patient underwent video assisted thoracotomy (VATS) of pleural exploration.

During the operation, a total of twenty-five smooth glossy white round-shaped, pearl-like lesions were observed in the pleura (Fig. 2A and B). The largest thoracolithiasis found in was 20mm. Most nodules were freely movable and discovered around the dependent position of the lung. There were some nodules that seemed to be originating from visceral & parietal pleura. There was no adhesion between the nodules and the organs around, and all the nodules were completely removed. Around the diaphragmatic pleura and nearby parietal pleura showed inflammatory change with minimal amount of serous effusion but otherwise no other abnormality was detected. Histopathological examination showed extensive necrotic fatty tissue at its center surrounded by fibrosis, as assumed from the thoracic image findings (Fig. 2C). The postoperative recovery was uneventful. The patient was symptom free after the surgical removal, suggesting thoracolithiasis

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Fig. 1. 39-year-old female with thoracolithiasis (A) An axial view of pre-contrast chest CT presenting chains of non-calcified well-defined nodules in the left hemithorax. Attenuation of nodules were 30–33 Hounsfield Units (HU). (B) In post-contrast chest CT, the nodules were not enhanced. (C) Thoracolithiasis on Chest MRI finding. (D) PET uptake was not observed.

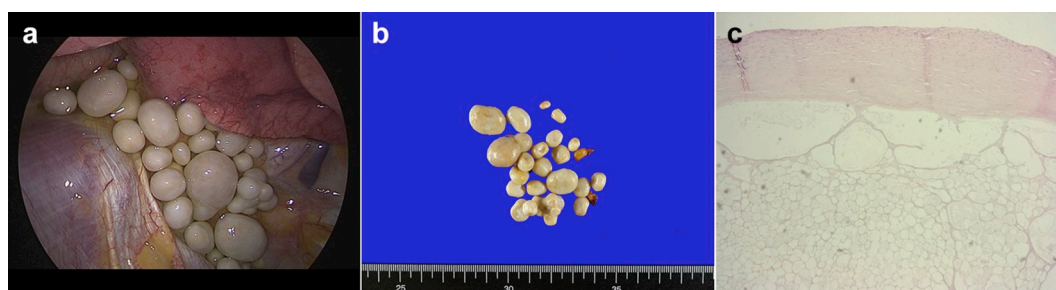


Fig. 2. (A) Intraoperation findings. (B) Gross specimen of multiple smooth glossy white round-shaped, various sized pearl-like lesion. (C) In high-power view of fat necrosis surrounded by fibrous tissue (hematoxylin-eosin stain, x40 magnification).

was associated with chest discomfort. She is still being followed up for about two years after the surgical removal with chest CT every year and the last CT scan showed no remarkable finding in the pleural cavity, lungs or mediastinum.

3. Discussion

Thoracolithiasis is a rare benign condition with a prevalence of 0.086% [2]. It was first reported as a “pleural stone” by Dias et al. [3] and is also named as pleural stone, or pleurolith, or intrathoracic calculus, or intrapleural loose body. There is no age or sex predilections [4] though a slight female dominance was observed in a previous report by Kinoshita et al. [2]. The size of thoracolithiasis ranges from 5 to 15 mm (1–3) and it may or may not be calcified. The histology has been described as an outer wall of fibrous tissue and a variable central core which is usually consisted of fatty tissue with or without necrosis [5]. In an asymptomatic patient, thoracolithiasis can be distinguished from other lesions by calcification, mobility, and an inferior intrapleural location on serial images. Mobility on sequential imaging especially is the most characteristic finding of thoracolithiasis [2,6,7].

When a thoracolithiasis is suspected, *trans*-thoracic or endobronchial biopsy often fails to obtain diagnosis because of its hard consistency and loose fixation within the pleural cavity [8]. With distinctive radiological findings, a major operation is not routinely needed nor recommended [9] especially in the absence of other evidence for neoplastic disease [2]. However in some cases, if it is non-calcified or enlarge at follow up studies or shows uncertain nature in images, then surgical removal should be considered for a definite diagnosis [1,3,10] Thoracoscopy under local anesthesia was reported to be minimally invasive and useful, and could be considered as an option for definite diagnosis [11].

The exact etiology of thoracolithiasis is unclear but few hypotheses considering its pathologic findings have been suggested. Pericardial fat necrosis tearing off in the pleural cavity, or old tuberculous foci, or an aggregation of macrophages phagocytizing dust may become round and polished after a long period associated with breathing [1,5,12]. Most of

thoracolithiasis have been found on the left side which is explained by the fact that the left side of thoracic cavity contains more pericardial fat than the right side.

Our patient had thoracolithiasis located in the dependent portion of the thorax, with non-calcified fibrosis with fat necrosis which support the pericardial fat necrosis theory. Comparing with typical findings from the previous cases [13], our patient was younger and had chest discomfort which was possibly resulted from the thoracolithiasis. She also had numerous thoracolithiasis which are distinctive findings. She underwent surgery because of her symptoms and the growing size at the follow up images. In a retrospective point of view, the nodules changed position in the left pleural and this mobile nature could have been a clue at initial diagnosis. Though rare entity, thoracolithiasis should be a different diagnosis, especially when a lesion is combined with mobility.

4. Conclusion

Due to its rare entity, thoracolithiasis is indistinguishable from a pulmonary nodule and may be under-recognized, while the mobility of the nodule is the key finding for the diagnosis.

Disclosure

The authors have no conflicts of interest to declare.

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