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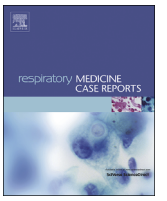

Case Report

Intrapulmonary solitary fibrous tumor with malignant potential: A case report

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ABSTRACT

Intrapulmonary solitary fibrous tumor is rare, and its clinical course has not been sufficiently reported. We presented a case of an 80-year-old male non-smoker and discussed the surgical procedure selection and the recurrence risk assessment. A solid nodule, 1.1 cm in diameter, was identified in the left lower lobe on chest computed tomography and showed no accumulation on positron emission tomography. A wedge resection with a sufficient surgical margin under video-assisted thoracoscopic surgery was performed. Based on histological morphology and immunohistochemical examination, this case was considered an intrapulmonary solitary fibrous tumor with malignancy potential, requiring cautious follow-up observation.

1. Introduction

A solitary fibrous tumor (SFT) is a ubiquitous mesenchymal tumor. In the thorax, 70 % of SFTs originate from the visceral pleura, and most others are derived from the mediastinal and parietal pleura [1]. Intrapulmonary SFT is extremely rare, and preoperative diagnosis poses a challenge. Moreover, some resected SFT cases have malignant potential and require a follow-up according to the recurrence risk. Therefore, selecting a surgical procedure with sufficient margin that preoperatively assumes intrapulmonary SFT is important. We describe a case of intrapulmonary SFT in which complete resection was achieved and discuss the surgical procedure selection and the recurrence risk evaluation.

2. Case presentation

An 80-year-old man, a non-smoker, was referred to us because of a nodule in the left lower lung on chest computed tomography (CT) during postoperative follow-up for squamous cell carcinoma of the right maxillary sinus (cT3N1M0 stage III), which had been treated with a multidisciplinary approach. Chest CT revealed a solid nodule measuring 1.1 cm in the peripheral region of the left lower lobe (Fig. 1A, B). Positron emission tomography (PET) showed no abnormal accumulation on the nodule (Fig. 1C).

Abbreviations: CT, computed tomography; HPF, high-power fields; PET, Positron emission tomography; SCC, squamous cell carcinoma antigen; SFT, solitary fibrous tumor.

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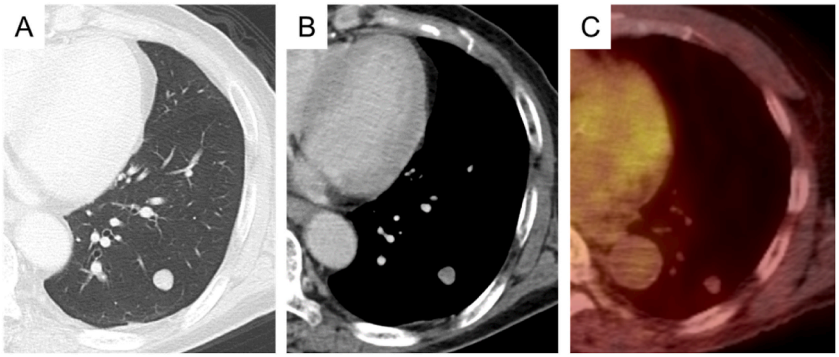

Fig. 1. Radiological findings. (A) Chest computed tomography showed a well-defined and rounded nodule in the left lower lung. (B) The nodule did not exhibit contrast enhancement and calcification. (C) Positron emission tomography revealed no accumulation in the lung nodule.

The serum level of squamous cell carcinoma antigen (SCC) was elevated at 6.6 ng/ml (normal value < 2.5 ng/ml). Based on a history of maxillary cancer and elevated SCC value, we preoperatively suspected a metastatic pulmonary tumor originating from the maxillary cancer. Subsequently, hybrid video-assisted thoracoscopic wedge resection of the left lower lobe was performed. The patient was discharged on postoperative day 6.

The tumor macroscopically presented as a white nodule with a well-defined margin (Fig. 2a). Histological examination revealed spindle cell proliferation with hypercellularity, separated from the visceral pleura (Fig. 2b and c). No nuclear atypia or necrosis was observed. There were four mitoses per 10 high-power fields (HPF) (Fig. 2d). Immunohistochemistry examination showed negativity for CAM5.2 (Fig. 2e), weak positivity for CD34 (Fig. 2f), positivity for signal transducer and activator of transcription 6 (Fig. 2g), and the Ki-67 labeling index of approximately 4 % (Fig. 2h).

Based on these findings, the patient was finally diagnosed with intrapulmonary SFT. The patient is undergoing follow-up for the recurrence of intrapulmonary SFT and maxillary cancer. One year after the surgery, no recurrence has been observed.

3. Discussion

Completely resected SFTs in the thorax typically have a favorable prognosis; however, recurrence occurs in 13–35 % of cases, leading to local recurrences at the surgical margin and distant metastatic recurrences [1–3]. The selection of a surgical procedure to prevent local recurrence and the assessment of risk to predict distant recurrence are important.

Complete resection with a sufficient margin for SFT is important because the primary form of recurrence in SFTs is local recurrence at the resection margin [4]. The surgical procedure for diagnosing and treating solitary pulmonary nodules is determined based on the preoperative diagnosis. However, preoperative diagnosis of intrapulmonary SFT is challenging because of the lack of distinctive imaging findings, making it difficult to distinguish from other diseases presenting as solitary pulmonary nodules. Generally, small intrapulmonary SFTs demonstrate well-defined nodules on CT and exhibit similar accumulation levels to the mediastinal blood pool on PET [5]. Differential diagnoses on radiological imaging include typical carcinoid and pulmonary hamartoma. The current case exhibited imaging characteristics consistent with intrapulmonary SFT. Based on the history of maxillary cancer and elevated serum SCC level, a metastatic pulmonary tumor originating from the maxillary cancer was suspected; however, the absence of abnormal accumulation on PET was inconsistent. Although the intraoperative appearance of the white, well-defined, and elastic tumor closely resembled a hamartoma, we did not select enucleation because of the lack of calcification within the nodule on preoperative imaging. Despite the difficulty in pre- and intra-operative diagnosis, we achieved complete resection with a sufficient margin, ensuring appropriate treatment. However, if a benign disease such as a pulmonary hamartoma were suspected, and a more localized wedge resection or tumor enucleation could be performed for diagnostic purposes; the risk of local recurrence might increase. Typically, intrapulmonary SFTs are centrally located, making it difficult to obtain sufficient surgical margins through wedge resection. Therefore, for solitary pulmonary tumors without characteristic radiological findings such as calcification, it is important to consider intrapulmonary SFT as a differential diagnosis based on preoperative imaging and determine the extent of lung resection such as segmentectomy and wide wedge resection for complete resection with sufficient margins.

SFT can develop distant metastatic recurrence. In intrapulmonary SFT, recurrent metastasis to the contralateral lung or chest wall, along with documented fatal cases, was reported [6]. Recurrent metastasis is more common in malignant SFT and is not curable [1]. Therefore, assessing malignancy is crucial for prognostic prediction. The four criteria for malignant SFT proposed by England et al. are well-known: (a) high cellularity with crowding and overlapping of nuclei, (b) pleomorphism based on nuclear size and irregularity, (c) nuclear polymorphism, and (d) more than 4 mitotic figures per 10 HPF [1]. This case would be considered to have malignant potential based on England's criteria due to hypercellularity [1]. Apart from England's criteria, several risk stratification models for malignant SFTs have been reported [7–9]. According to these models, hypercellularity and 4/10 HPF have been identified as predicted factors for malignant SFT. Additionally, risk factors such as an elevated maximum standardized uptake value on PET [10], Ki-67 LI > 10 % [3], and decreasing expression of CD34 [11], as observed in this case, have been reported. Therefore, this case should be noted for distant recurrence more than benign cases. Moreover, recurrence more than 5 years after surgery has been reported in in-

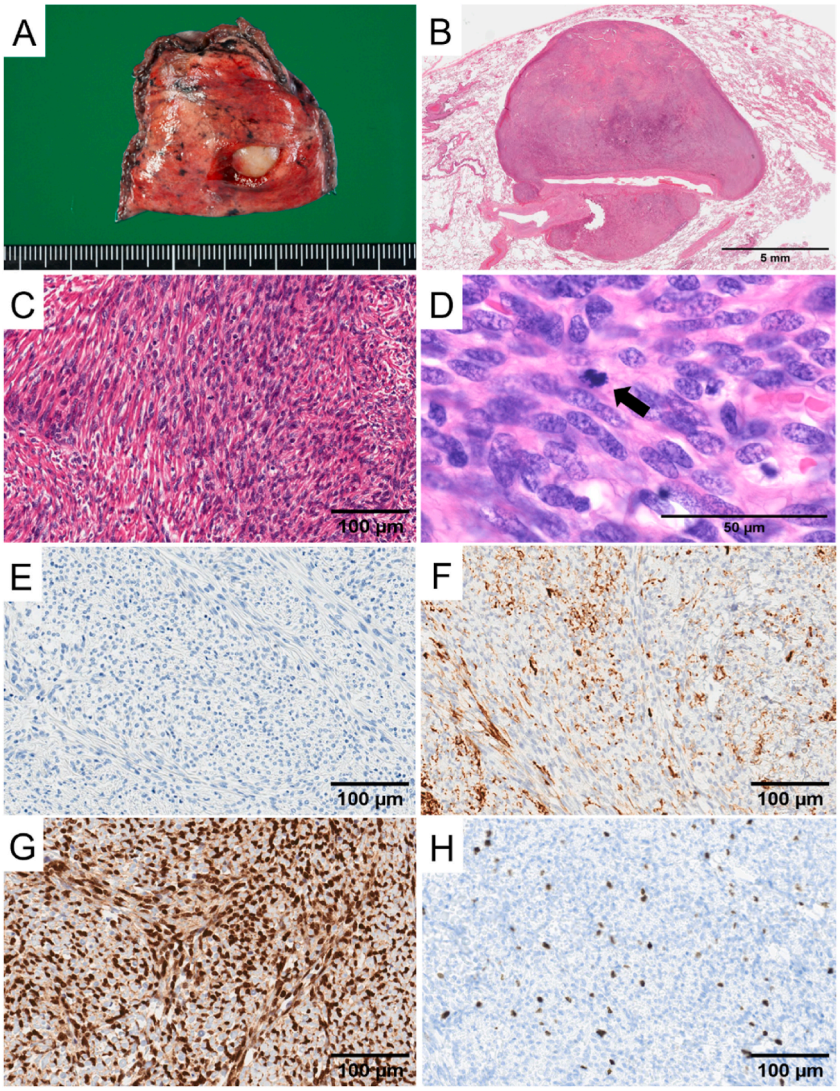

Fig. 2. Histopathological findings of the tumor in the left lower lung. (A) The tumor was a white nodule with a well-defined margin. (B)(C) The hematoxylin-eosin stain demonstrated spindle cell proliferation within the lung parenchyma, separated from the visceral pleura. (D) Scattered mitosis was observed (arrow). In immunohistochemical examinations, (E) CAM5.2 was negative, (F) CD34 was weakly positive, (G) signal transducer and activator of transcription 6 was positive, and (H) the Ki-67 labeling index was 4 %.

trapulmonary SFTs [6,12]. Long-term and careful follow-up observation should be required for intrapulmonary SFT with malignant potential.

4. Conclusion

Intrapulmonary SFT is rare and lacks characteristic imaging findings, making preoperative diagnosis challenging. Suspecting this disease as a differential diagnosis and selecting a surgical procedure with sufficient surgical margins are important for preventing local recurrence. Additionally, it is necessary to assess the malignancy comprehensively by considering risk scores and risk factors and to maintain cautious postoperative follow-up observation.

Author contribution statement

Takamitsu Hayakawa drafted the manuscript. Yusuke Takanashi, Yuta Matsubayashi, Keigo Sekihara, Akikazu Kawase, and Kazuhito Funai collected clinical data, commented on and revised the manuscript. Mana Goto and Satoshi Baba diagnosed pathologically, commented on and revised the manuscript. Tomoya Tajiri, Motohisa Shibata and Norihiko Shiiya commented and revised the manuscript. All authors approved the final manuscript critically.

Ethics statement

The authors declare that appropriate written informed consent was obtained for the publication of this manuscript and accompanying images.

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Declaration of competing interest

No conflict.

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