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A primary cellular fibrohistiocytoma of the lung: Report of a case

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ABSTRACT

INTRODUCTION: Cellular fibrohistiocytoma (CFH) is a type of fibrohistiocytic tumor that commonly occurs in the dermis and superficial subcutis. The designation is used for lesions that show increased cellularity with a fascicular growth pattern and frequent extension. Our search of literature only revealed one case of a primary CFH of the lung. We experienced a rare patient with a primary CFH of the lung.

PRESENTATION OF CASE: We herein present a rare case of a 77-year-old female patient without a cutaneous lesion, who underwent resection for what was considered to be a primary CFH of the lung. There has been no recurrence including a cutaneous lesion in a year after surgery.

DISCUSSION: CFH is considered to be benign, but rare cases showing multiple recurrences and involving metastasis to the lymph nodes and internal organs have been reported. At present, it is not possible to predict this aggressive biological behavior based on the tumor histology.

CONCLUSION: It is essential to perform resection with an adequate margin with close clinical follow-up.

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1. Introduction

Benign fibrous histiocytoma is a type of fibro-histiocytic tumor that commonly occurs in the dermis and the superficial subcutis. The tumor is characterized by the tumoral differentiations of fibroblasts and histiocytes, the etiology of which is unknown. Cellular fibrous histiocytoma (CFH), which was first described by Calonje et al., is a distinct variant of fibrous histiocytoma. CFH accounts for approximately 5% of cutaneous benign fibrous histiocytomas [1]. In contrast to ordinary fibrous histiocytoma, CFH displays high cellularity, limited polymorphism, a fascicular growth pattern and a partly storiform pattern. Although it is generally considered to be a benign tumor, CFH shows a tendency for local recurrence. We herein present a rare case of a patient with a primary CFH of the lung.

Abbreviations: CFH, cellular fibrous histiocytoma; CT, computed tomography; VATS, video-assisted thoracoscopic surgery; DFSP, dermatofibrosarcoma protuberans.

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2. Case report

The patient was a 77-year-old asymptomatic female who visited our hospital after an abnormal shadow was detected on a chest X-ray. She had no history of cutaneous lesions nor did she have any history of occupational exposure to silica, beryllium, or asbestos. Chest computed tomography (CT) revealed the presence of a well-demarcated solid nodule of 2.3 cm in diameter in the right lower lobe (Fig. 1). A systemic CT examination revealed no tumors other than this pulmonary tumor.

We performed a wedge resection of the right lower lobe by video-assisted thoracoscopic surgery (VATS). Three ports were placed in the left lateral decubitus position. The lung was deflated to confirm the right lung nodule. A linear stapling device was used to resect the lung nodule. The nodule was firm and solid. On sectioning, it appeared yellow-white in color (Fig. 2). A frozen section revealed the proliferation of fibromuscular tissue with a few mitotic figures, suggesting that it was a mesenchymal tumor. Right lower lobectomy was performed because malignancy could not be denied based on the examination of the frozen section. The postoperative course was uncomplicated. The histopathological findings revealed a well-demarcated nodular lesion with the cellular proliferation of almost bland-looking spindle or oval histiocytoid cells, arranged in a storiform or loose fascicular fashion with many branching or anastomosing blood vessels, sometimes displaying hemangiopericytomatous pattern, in the pulmonary parenchyma. Mitotic figures

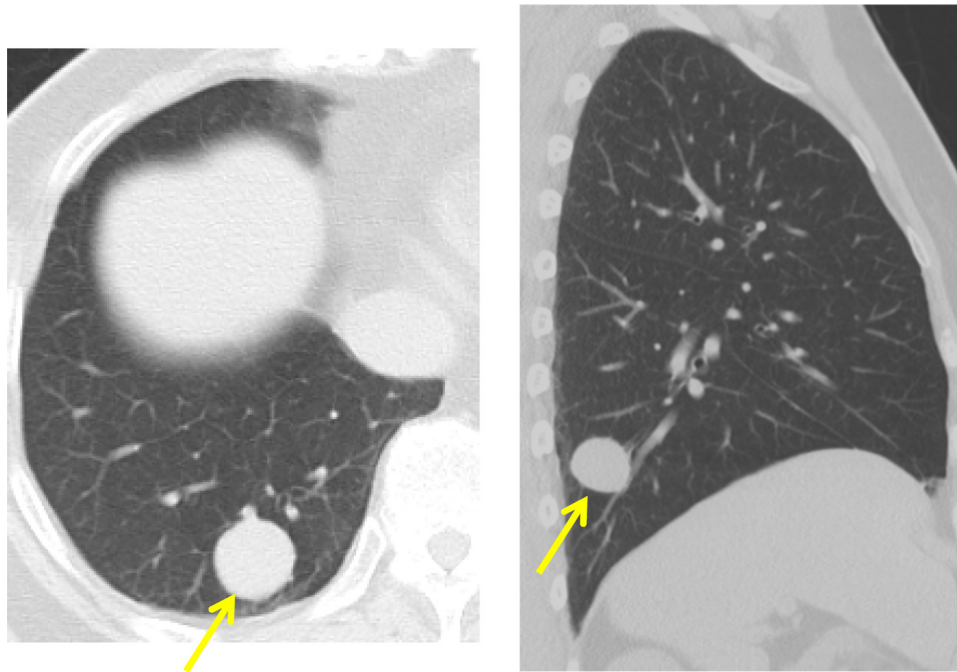


Fig. 1. Chest computed tomography (CT) revealed the presence of a well-demarcated solid nodule of 2.3 cm in diameter in the right lower lobe.

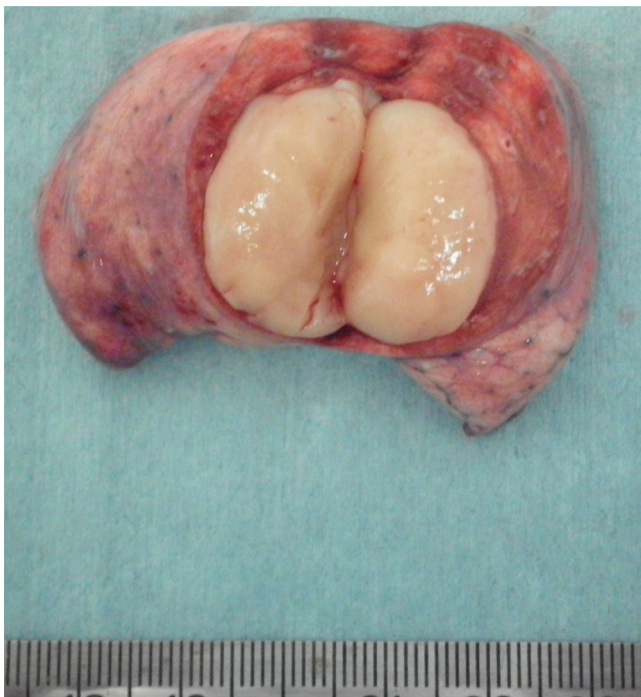


Fig. 2. A nodule was firm, solid, and yellow-white in color on sectioning.

were sometimes encountered (Fig. 3A). Immunohistochemically, many tumor cells were positive for alpha-smooth muscle actin (Fig. 3B), estrogen receptor and progesterone receptor and some were also positive for bcl-2 and CD10. A small number of factor XIIIa-positive cells were admixed. The specimens were negative for CD34, CD68, desmin, cytokeratins (AE1/AE3, CAM5.2), EMA, h-caldesmon, transgelin, CD99, TLE-1 and STAT6 (Fig. 3). In our molecular assay using formalin-fixed, paraffin-embedded tumor tissue specimens, no S18-SSX fusion gene transcript was detectable, despite the use of amplifiable PBGD and PGK gene transcripts as internal controls. The lesion was diagnosed based on the features,

which were suggestive of CFH. There has been no recurrence including a cutaneous lesion in a year after surgery and the patient's condition has remained good.

3. Discussion

CFH represents a morphological variant of fibrous histiocytoma, which should be distinguished from dermatofibrosarcoma protuberans (DFSP) and leiomyoma. Microscopically, CFH shows high cellularity and is composed of spindle-shaped cells with variable amounts of eosinophilic cytoplasm and small, oval, vesicular eosinophilic nuclei. The histological findings of CFH include large size, higher cellularity with a more fascicular architecture, a focally smooth muscle-like appearance, a moderate mitotic rate, focal areas of necrosis or infarction, and limited cellular polymorphism. Cytological atypia is not a feature. The tumors include lymphocytes, plasma cells, foamy macrophages and less commonly, giant cells. Most of these findings are seen in the cases that we have treated and these microscopic findings are thought to be imperative for making a conclusive diagnosis, as we did in the present case. Traditionally, CD34 and factor XIIIa have been widely used in distinguishing CFH from DFSP [2]. Although CD34 expression was not observed in the present case, CD34 is observed in up to 20% of CFH cases [2]. Even in the subset of CFH patients in whom CD34 positivity is not observed, the tumor can be correctly diagnosed based on the characteristic morphological features, which readily distinguish CFH from DFSP.

Most CFHs occupy the superficial dermis and extend into the deep reticular dermis. In this case we diagnosed the lung lesion as a primary CFH of the lung because no other lesions had been detected in her body in a year after surgery. It should be noted, however, that pulmonary CFH lesions are often metastases [3,4]. Our search of literature only revealed one case of primary CFH of the lung. An 8-year-old boy underwent right middle and lower lobectomy after a $4.0 \times 4.0 \times 3.0$ cm tumor was located in the median of the bronchus intermedius. Microscopically, the lesion showed high cellularity, limited polymorphism, a fascicular growth pattern and a partly storiform pattern [5].

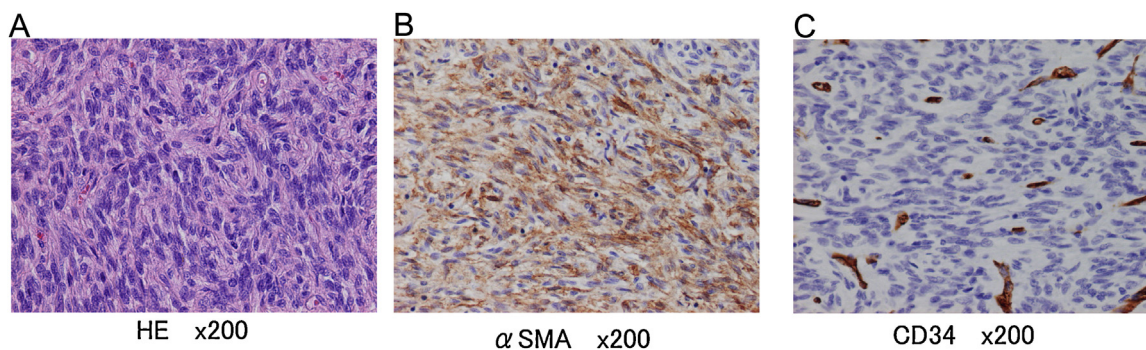


Fig. 3. (A) The histopathological findings revealed a well-demarcated nodular lesion with the cellular proliferation of almost bland-looking spindle or oval histiocytoid cells, arranged in a storiform or loose fascicular fashion with many branching or anastomosing blood vessels, sometimes displaying hemangiopericytomatous pattern, in the pulmonary parenchyma. Mitotic figures were sometimes encountered ($\times 200$). (B) Immunostaining for alpha-smooth muscle actin showed that many tumor cells expressed alpha-smooth muscle protein ($\times 200$). (C) Immunostaining for CD34 showed that tumor cells expressed no CD34 protein ($\times 200$).

Surgical intervention is important, not only for achieving a cure, but also as a diagnostic measure to rule out malignancy. Complete resection alone seems to be a suitable surgical treatment. There is increasing evidence to suggest that patients with CFH experience local recurrence more frequently than those with usual fibrous histiocytomas (in whom the recurrence rate is 25%), especially after incomplete surgical excision [6]. CFH is considered to be benign, but rare cases showing multiple recurrences and involving metastasis to the lymph nodes and internal organs have been reported [4,7]. At present, it is not possible to predict this aggressive biological behavior based on the tumor histology. It is therefore essential to perform resection with an adequate margin with close clinical follow-up.

We herein presented a rare resected case of a primary CFH of the lung and discussed its differential diagnosis and histogenesis.

Conflict of interest

None.

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None.

Ethical approval

None.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in Chief of this journal on request.

Author contribution

Yoshinobu Ichiki: study design, data collections, data analysis, writing.

Junji Kawasaki : data collections.

Takayuki Hamatsu: data collections.

Taketoshi Suehiro : data collections.

Fumihiko Tanaka: study design, data collections, data analysis.

Mansanori Hisaoka: study design, data collections, data analysis.

Keizo Sugimachi: data collections.

Guarantor

Yoshinobu Ichiki.

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