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CASE REPORT

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Multiple calcifying fibrous tumor of the pleura: A case report

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Abstract

Calcifying fibrous tumor of the pleura (CFTP) is a rare benign tumor of the thoracic cavity. Due to the low incidence of CFPT, it is prone to be misdiagnosed because intraoperative analysis of frozen section is a challenge for pathologists. At present, it is difficult to distinguish this tumor from other benign thoracic tumors based on radiographic features. Therefore, surgical resection is the best method for definite diagnosis and treatment.

KEYWORDS
benign tumor, calcifying fibrous tumor, pleura

INTRODUCTION

Calcifying fibrous tumor (CFT), a rare benign tumor originally reported by Rosenthal and Abdul-Karim in 1988,[1] occurs in many parts of the body, including the subcutaneous soft tissue, gastrointestinal tract, and pleura. In 2002, the World Health Organization (WHO) established the name as "calcifying fibrous tumor" in the classification of tumors of soft tissue and bone.[2] Approximately 10% of CFT cases have been reported in the pleura.[3] Calcifying fibrous tumor of the pleura (CFTP) was first described in 1996 by Pinkard et al.[4] We present the case from a 38-year-old male with multiple CFP of the pleura, and perform a literature review of pleural CFT.

CASE REPORT

A 38-year-old man was admitted to our hospital with intermittent right chest pain. Computed tomography (CT) scan of the chest incidentally discovered multiple soft tissue masses within the right basilar pleura and the largest node was 5.0 cm in maximum diameter. There was associated mild right pleural thickening with a small pleural effusion. Tumor marker associated with lung cancer was negative. Tumor positron emission tomography (PET) and CT imaging using fluorodeoxyglucose F18 (F18-FDG) revealed FDG accumulation and a maximum standardized uptake value of 1.8 in the tumor(Figure 1). As we were not able to diagnose the tumor using a CT-guided needle biopsy, the patient underwent an excisional biopsy via right video-assisted thoracic surgery to confirm the diagnosis. The procedure identified multiple firm, pearly white masses on both the visceral and parietal pleura, including the diaphragm, and multiple small nodules were near the largest mass located in right lower lobe (Figure 2). As the intraoperative frozen pathological analysis was considered to be mesenchymal tumor accompanied by a large number of inflammatory lymphocytic infiltration, incomplete resection was performed. Postoperative paraffin section pathology indicated that the lesion was relatively well-circumscribed and non-capsulated, composed of a large number of dense collagen fiber hyperplasia and minute psammomatous calcifications, and the tumor was well defined from lung tissue and was composed of fibrous connective tissue rich in collagen (Figure 3). The tumor consisted of spindle cells in which apparent nuclear atypia, fission image, and necrosis were not observed, and scattered calcification or gravel formation can be seen (Figure 4) and scattered positive for CD34 and STAT6, but negative for CK7, CK5/6, CD68, and S100. Based on the histologic and immunohistochemistry findings, a diagnosis of CFP was made.

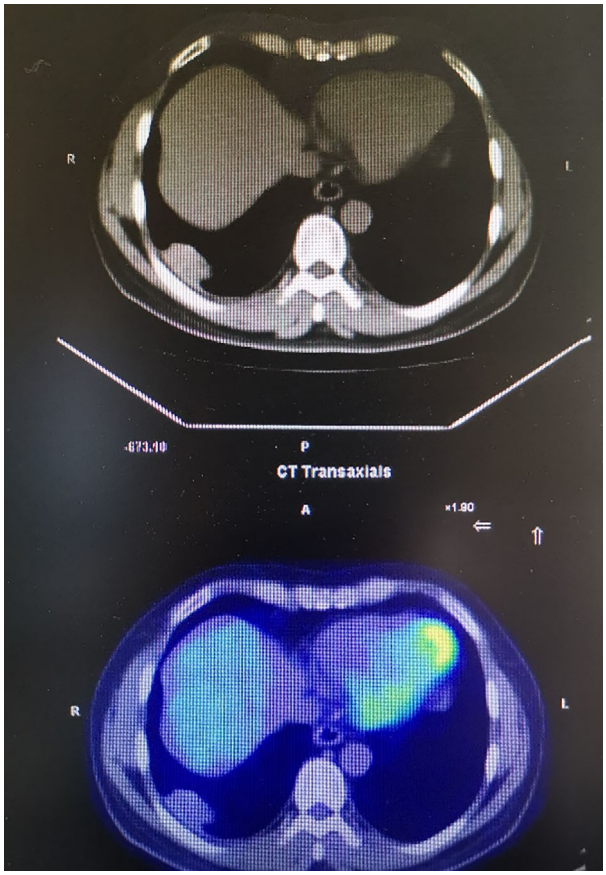

FIGURE 1 Computed tomographic scan reveals a subpleural mass with dystrophic calcification in the right lower thoracic cavity

FIGURE 2 The largest mass was on the surface of right lower lung and wedge resection was performed

DISCUSSION

CFT was once called "calcifying fibrous pseudotumor". In 2015 World Health Organization classification of lung and pleural tumors, this lesion has been renamed calcifying fibrous tumor rather than pseudotumor because of its tendency to local recurrence.[5] We reviewed the literature on CFTP in both English and non-English, identifying 32 total

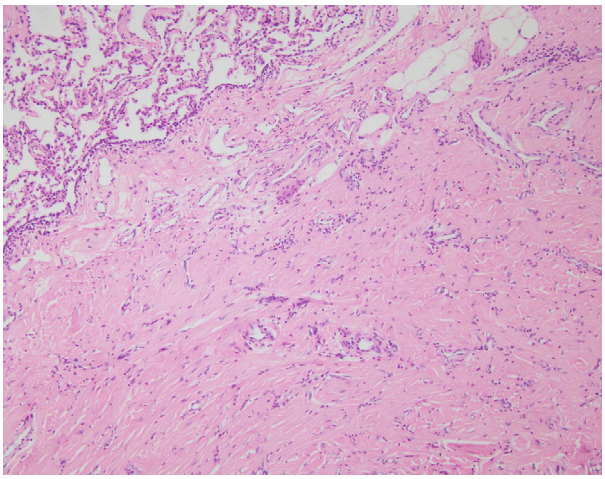

FIGURE 3 The microscopic feature of the tumor is well defined from lung tissue and is composed of fibrous connective tissue rich in collagen (hematoxylin & eosin stain, ◊100)

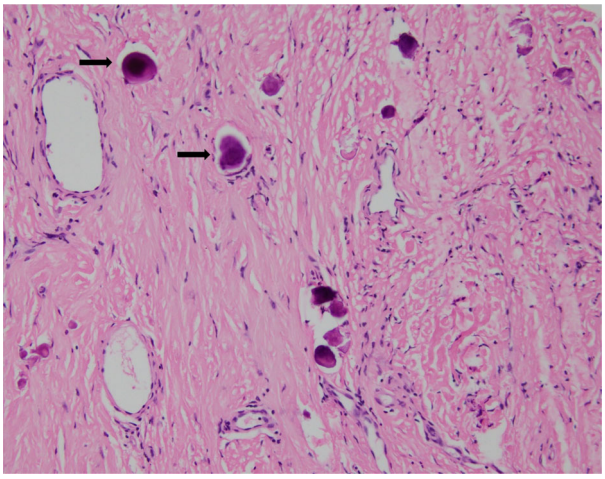

FIGURE 4 Scattered calcification or gravel formation in tumor (hematoxylin & eosin stain, ◊200)

cases' including our own (Table 1).[4,6–32] Patients had an average age of 34.1 years (range 7–59), 53% were female and 71.9% (23/32) showed multifocal pleural disease. Therefore, CFTP mostly occurs in younger patients and has multiple lesions.

CFTP can be asymptomatic for many years before presenting with symptoms mostly located in the lower thoracic cavity, rarely involving the apical pleural surfaces.[30] In our case, the lesions were located in the right lower lobe, diaphragm, and right costophrenic angle. CFTP needs to be differentiated from solitary fibroma (SFT), inflammatory myofibroblastoma (IMT), malignant pleural mesothelioma, chest wall sarcoma, calcified pleural plaque, and chronic reactive pleurisy. As it is difficult to distinguish these diseases based on imaging, definitive diagnosis mainly rely on histological and immunohistological assessments. CFTP is

TABLE 1 Reported cases of pleural calcifying fibrous tumor

Case	Author [ref no.[4,6-32]] (year)	Age/gender	Focality	Surgical resection
1	Pinkard et al. (1996)	23/F	Multiple	Complete
2	Pinkard et al. (1996)	28/F	Multiple	Complete
3	Pinkard et al. (1996)	34/M	Solitary	Complete
4	Hainaut et al. (1999)	29/F	Multiple	Incomplete
5	Cavazza et al. (2002)	46/F	Solitary	Complete
6	Ammar et al. (2003)	38/F	Solitary	Complete
7	Jang et al. (2004)	31/F	Solitary	Complete
8	Soyer et al.(2004)	7/M	Solitary	Complete
9	Mito et al. (2005)	54/M	Multiple	Incomplete
10	Kawhara et al. (2005)	35/F	Multiple	Incomplete
11	Shibata et al. (2008)	54/F	Multiple	Incomplete
12	Suh et al. (2008)	35/M	Multiple	Complete
13	Miyano et al. (2008)	44/F	Multiple	Complete
14	Sleigh et al. (2009)	22/F	Multiple	Incomplete
15	Chang et al. (2009)	37/M	Multiple	Complete
16	Isaka et al. (2010)	40/M	Multiple	Complete
17	Jiang et al. (2010)	44/F	Multiple	Complete
18	Ağaçkıran et al. (2012)	40/M	Multiple	Complete
19	Ishida and Okabe (2013)	31/M	Multiple	Incomplete
20	Azam et al. (2014)	31/M	Multiple	No
21	Minerowicz et al. (2015)	15/F	Multiple	Incomplete
22	Lee et al. (2015)	47/F	Solitary	Complete
23	Rocas et al. (2015)	59/M	Solitary	Complete
24	Mazi et al. (2017)	15/F	Multiple	Incomplete
25	Lisowska et al. (2018)	27/M	Solitary	Complete
26	Mehrad et al. (2018)	32/M	Multiple	Incomplete
27	Mehrad et al. (2018)	21/M	Multiple	Complete
28	Mehrad et al. (2018)	32/F	Solitary	Complete
29	Edlin et al. (2018)	23/F	Multiple	Complete
30	Massoth et al. (2019)	59/M	Multiple	Incomplete
31	Miyamoto et al. (2020)	21/F	Multiple	Incomplete
32	Hernandez et al. (2020)	35/M	Multiple	Complete
33	Current case (2021)	38/M	Multiple	Incomplete

benign and multifocal, and it is recommended to remove all nodules as far as possible. Due to the lack of long-term follow-up data for incomplete resection cases and no definitive data on postoperative recurrence, it is not yet proven that the prognosis of patients with partial resection is worse than that of patients with complete resection. Currently, the pathogenesis of CFPT is not clear. Chorti et al.[3] considered the possibility of genetic alterations or perhaps an embryologic factor. Mehrad et al.[28] recently found deleterious mutations in three genes, ZN717, FRG1, and CDC27, as well as abnormal copy number losses on chromosome 8 and 6 by whole-exome sequencing in three CFPT patents, suggesting that these molecular level changes may contribute to CFTP tumorigenesis. There is debate as to whether CFPT is a multisource lesion or whether it spreads from the main lesion to nearby pleura; the exact mechanism underlying this dissemination is unclear. Massoth et al.[30] reported that reactive-appearing adhesions involved by CFPT may be the mode of dissemination across the pleural surfaces. In our case, we did not find the "reactive-appearing adhesions" described by Massoth. Therefore, the mechanism of tumor involving adhesions needs to be verified by subsequent research. Due to the low incidence of CFTP, large sample studies are impossible. Therefore, every case of CFPT should be reported to facilitate further understanding of its pathogenesis and dissemination mechanism.

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