Case Report

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Multiple pulmonary hyalinizing granulomas mimicking widespread lung metastasis

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Abstract:

Pulmonary hyalinizing granuloma (PHG) is a rare benign lesion that can be unilaterally or bilaterally presented in the form of solitary or multiple nodules and masses. It may also be related to mediastinal and retroperitoneal fibrosis, autoimmune diseases, tumors, or infectious diseases. The definitive diagnosis of PHG can only be made by pathological evaluation. A 64-year-old male applied to the hospital to undergo an inguinal hernia repair operation. Preoperative chest imaging revealed multiple pulmonary nodules with lobulated contours and regular margins scattered throughout both lungs. Pathologic evaluation revealed that nodules were consistent with PHG. In addition, soft-tissue density observed around the abdominal aorta was compatible with retroperitoneal fibrosis, which may accompany this disease.

Keywords:

Computed tomography, lung nodule, pulmonary hyalinizing granuloma, retroperitoneal fibrosis

Introduction

Pulmonary hyalinizing granuloma (PHG) is a rare benign lesion that can be unilaterally or bilaterally presented in the form of solitary or multiple nodules and masses. It is a rare fibrosclerosing inflammatory lung disease of unknown cause. Two hypotheses have been suggested as the potential pathogenic mechanism: an autoimmune process and an end-stage inflammatory process.[1] It may be related to mediastinal and retroperitoneal fibrosis, autoimmune diseases, or infectious diseases.[2] The definitive diagnosis of PHG can only be made by pathological evaluation. Histologically, it is characterized by lymphocyte and plasma cells surrounding vitrified collagen fibers. In this case report, we presented a patient who has multiple pulmonary nodules imitating metastatic lung disease and was diagnosed with PHG after surgery.

Case Report

A 64-year-old male applied to the hospital to undergo an inguinal hernia repair operation. In the physical examination, except for mild chest pain, the patient had no active complaints. He had no history of prior lung disease or malignancy. Preoperative chest radiography demonstrated smoothly defined pulmonary nodules in the left lung [Figure 1]. Chest computed tomography (CT) of the patient revealed multiple pulmonary nodules with lobulated contours and regular margins scattered throughout both lungs,

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the largest measuring 33 mm × 26 mm in size located in the lingula of the left lung [Figures 2 and 3]. In addition, homogeneous soft-tissue density surrounding the abdominal aorta was observed [Figure 4]. There was no mediastinal or abdominal lymphadenopathy. It was decided to biopsy with video-assisted thoracoscopic surgery from the nodule located at the lingula of the left upper lobe [Figure 3]. Pathologic evaluation revealed that nodules consist of thick and lamellar collagen fibers arrayed in a whorled or concentric pattern around small blood vessels that were characteristic for PHG [Figures 5 and 6]. Mononuclear inflammatory cells were seen, especially at the periphery of the lesions [Figure 6]. Large vessels detected in the lesion were obliterated and inflamed [Figure 6]. Immunostaining that was performed for plasma cell, IgG, and IgG4 did not reveal any specific finding for IgG-related disease.

Discussion

PHG is a benign neoplastic lesion and is mainly characterized by rich fibrosis. No apparent age, gender, or racial domination has been identified in the literature. The disease is generally known to be asymptomatic unless there is not a large number of lesions. Still, as in our case, dyspnea may occur when there are multiple lesions with progressive enlargement.[3] The other common symptoms are cough, and chest pain.[4,5] PHG has been thought to be associated with mediastinal and retroperitoneal fibrosis, as well as autoimmune, hematologic, thromboembolic, and infectious diseases.[1,6] In our patient, soft-tissue density observed around the abdominal aorta was compatible with retroperitoneal fibrosis, which may accompany this disease. Although autoimmune diseases may also be associated with PHG, antinuclear antibody, antineutrophil cytoplasmic antibody, and other autoantibodies were negative in our patient, showing that he did not show any signs of collagen tissue disease.

PHG lesions are often located in the subpleural pulmonary parenchyma. The calcified masses are more often multiple and bilateral in PHG.[1] Cavitation is not an expected condition but has been reported in a few cases.[1,7] The nodule size varies from a few millimeters to 15 cm, with an average of 2 cm.[8] However, in this case,

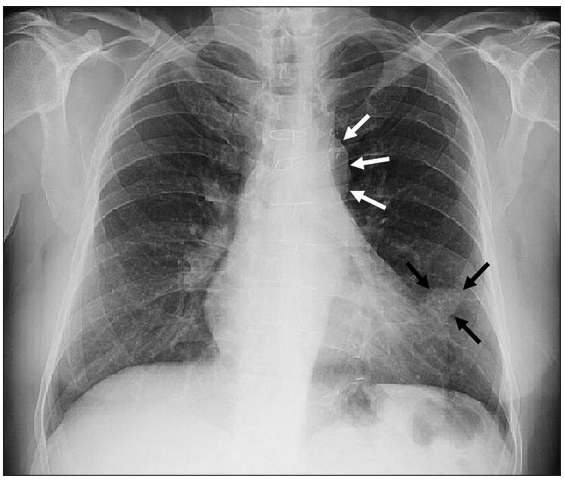

Figure 1: Chest X-ray of the patient shows a centrally located mass above the left hilum (white arrows) and another smoothly defined nodule at left lower zone adjacent to the left heart border (black arrows)

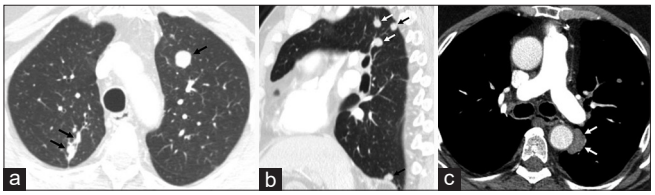

Figure 2: Bilateral multiple solid pulmonary nodules with regular margins are detected in the computed tomography of the patient. (a) Axial computed tomography image in the lung window shows bilateral pulmonary nodules of different sizes. (b) Sagittal CT image in the lung window shows multiple well-marginated pulmonary nodules. (c) Axial CT image in the mediastinal window shows solid pulmonary nodule with lobulated contours adjacent to the aorta

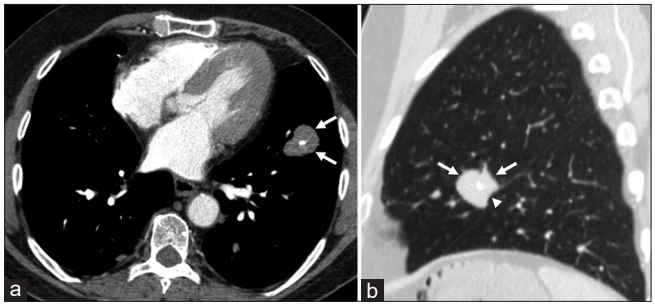

Figure 3: (a) Axial computed tomography image in the mediastinal window showing a well-marginated and lobulated mass with central calcification at the lingula of the left upper lobe. (b) Sagittal computed tomography image in the lung window shows retraction of the fissure (arrowhead) caused by the nodule located in the left upper lobe

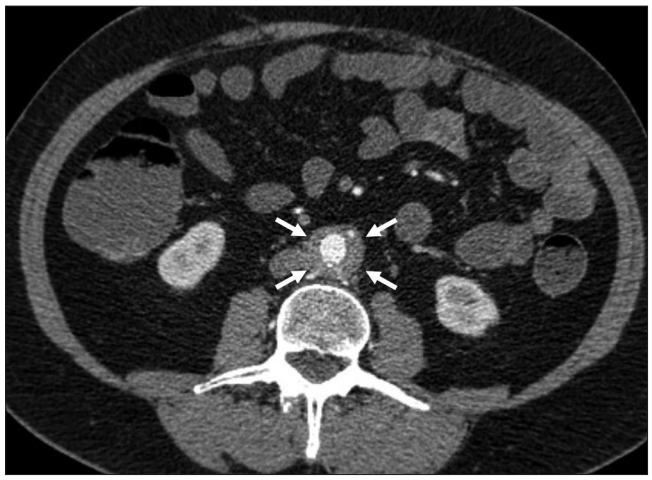

Figure 4: Axial enhanced computed tomography image shows soft tissue density around the abdominal aorta, compatible with retroperitoneal fibrosis, which may accompany pulmonary hyalinizing granuloma

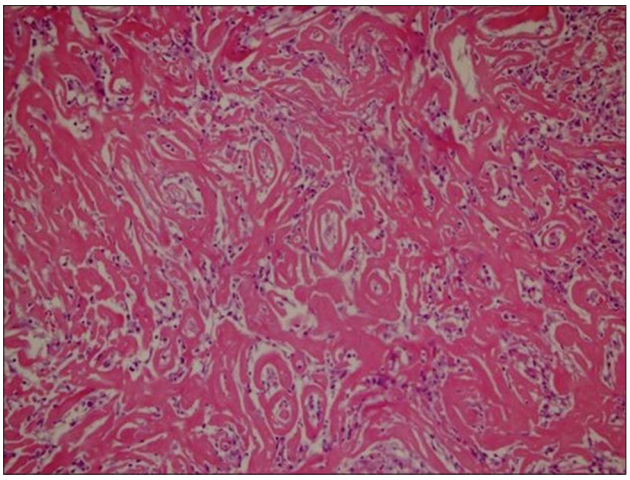

Figure 5: The characteristic ropy or lamellar hyaline collagen fibers that arrayed in a whorled or storiform pattern at high magnification (H and E, ×20)

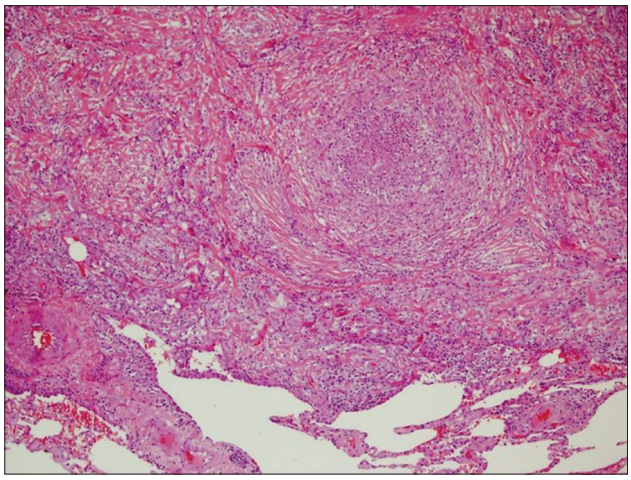

Figure 6: Associated chronic inflammatory infiltrates and obliterated blood vessels at high magnification (H and E, ×10)

the longest diameter of the lesion was 33 mm, which is above the average. Lymphadenopathy is not an expected condition in PHG,[3] as in our patient.

The differential diagnosis of multiple pulmonary nodules with regular borders should suggest metastasis, sarcoidosis, rheumatoid nodules, granulomatous polyangiitis, tuberculosis, amyloidosis, and lung cancer.[1,9] In the case of multiple calcified nodules, the differential diagnosis should include sarcomatous metastases (e.g., osteosarcoma, chondrosarcoma, and giant cell tumor) and carcinomatous metastases (e.g., mucin-producing adenocarcinomas, thyroid cancer, and choriocarcinoma).[1] However, our patient had no history of malignancy, tuberculosis, or any other prior lung disease.

It is not possible to make a definitive diagnosis of PHG radiologically and differentiate it from primary lung cancer or metastasis based on radiological findings. Both PHG and lung cancer can be seen as solitary or multiple nodules with well-defined or irregular borders. Therefore, histopathological evaluation is essential for making a definitive diagnosis. It has been reported that bronchoalveolar lavage and transbronchial biopsy are insufficient for the diagnosis of PHG.[10] Consequently, CT-guided percutaneous biopsy or surgical resection is needed to confirm the diagnosis.

The prognosis of PHG is generally good. While surgical resection is therapeutic in the presence of a single nodule, this is not possible for multiple nodules. There are some studies suggesting glucocorticoid therapy for multiple PHG; however, the effectiveness of this approach is still unclear.[10]

In conclusion, PHG is a benign lesion that should be included in the differential diagnosis of pulmonary nodules and masses. It may be considered in the differential diagnosis, especially in cases with no history of primary malignancy or lung cancer but who has autoimmune, hematological, thromboembolic, and infectious diseases or mediastinal and retroperitoneal fibrosis, as in our case.

Informed consent

Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understand that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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