

Case Report

Multiloculated Cavitory Primary Pulmonary Hodgkin Lymphoma: Case Series

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Keywords

Cavitory lesion · Hodgkin lymphoma · Primary pulmonary Hodgkin lymphoma

Abstract

Primary pulmonary Hodgkin lymphoma (PPHL) is very rare and typically involves the superior portion of the lung. Pulmonary involvement is observed in 15–40% of Hodgkin lymphoma patients. Three such patients who presented with an unusual form of PPHL in radiological studies, i.e., multiloculated cavitory lesions, were admitted to our hospital. These lesions represent a new pathological and radiological feature of PPHL.

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Introduction

Primary pulmonary Hodgkin lymphoma (PPHL) is a rare tumor, but reports of this malignancy have become more frequent in recent years. Pulmonary manifestations of Hodgkin lymphoma (HL) are usually characterized by a single nodule, multiple nodules, a single cavitory lesion or a mass. However, the patients described herein presented with multiloculated cavitory lesions, which are a very rare radiological manifestation. The aim of this report is to add to the current literature and examine possible correlations with other differentials in this geographical area.

Case 1

A 16-year-old male presented with fever, weight loss, and a productive cough for several weeks. On examination, his temperature was 39.5°C, and decreased air entry and fine crepitation in the right middle zone were observed. The laboratory tests indicated a white blood cell (WBC) count of 35,000/μL. His erythrocyte sedimentation rate (ESR) was 120 mm/h and his C-reactive protein (CRP) level was 15 mg/dL. The blood culture and acid fast bacilli (AFB) stain were negative. A chest X-ray (CXR) showed a large cavity lesion with a smooth margin and a thickened wall with air fluid levels associated with adjacent lung consolidation that involved the right middle and lower lung zones silhouetting the cardiac border. An enhanced chest CT scan showed a large multiloculated cavitory lesion that involved the right middle lobe and the anterior mediastinum, with internal air and fluid. Some reactive mediastinal lymph nodes were observed (Fig. 1). Video-assisted thoracoscopy (VATS) was performed due to the inconclusive nature of the CT-guided biopsy. Bronchoalveolar lavage (BAL) cytology was negative for malignant cells. The histopathology was consistent with classic HL nodular sclerosing type disease, and the immunohistochemistry was positive for CD30, CD68, CD3, CD20, and LCA but negative for CD15 and CK.

The patient was referred to an oncology service for further management.

Case 2

A 29-year-old female diagnosed with bronchial asthma presented with a dry cough that converted to a purulent productive cough for several weeks. She subsequently experienced episodes of hemoptysis. Her physical examination was unremarkable. Her initial WBC count was 8.4 k/μL. ESR was 84 mm/h and her CRP was 5.5 mg/dL. A septic workup indicated that all cultures and an AFB stain were negative. Her CXR showed a cavitory lesion involving the right middle zone of the lung. An enhanced chest CT scan showed a 4.4 × 4.6 × 6.3 cm mass with central hypodensity indicating a multiloculated cavitory lesion, and air was observed at the right anterior mediastinum and anterior segment of the right upper lobe. Moreover, enlarged reactive lymph nodes were observed along the prevascular space and para-aortic region (Fig. 2a, b). The patient underwent a CT-guided biopsy, and the histopathology was

consistent with nodular sclerosing type HL. The immunohistochemistry was positive for CD30, CD15, and CD3 with CD20 lymphocytes.

The patient was referred to an oncology service for further management.

Case 3

A 21-year-old male diagnosed with hypothyroidism and sickle cell anemia presented with fever and a productive cough for 4 months. He indicated a history of night sweats and weight loss. On examination, right anterior 3 × 3 cm cervical swelling was palpable. The thyroid gland was slightly enlarged. A chest examination demonstrated bilateral decreased air entry over the middle and lower zones. His initial WBC count was 14.2 k/μL. His CRP was 5.8 mg/dL and his ESR was 120 mm/h. A septic workup indicated that all cultures and AFB staining were negative. The CXR showed a diffuse heterogeneous opacity in the right middle zone. An enhanced chest CT scan showed a large mass extending from the anterior aspect of the mediastinum with an intrapulmonary component measuring 11 × 9 × 12 cm and a necrotic center. The thick enhancing soft tissue rim indicated a multiloculated cavitory lesion (Fig. 3a, b). Moreover, the echocardiogram showed pericardial effusion. The CT-guided biopsy was consistent with classic nodular sclerosing type HL, and the immunohistochemistry was positive for CD30 and CD15 but negative for CD45, CD3, and CD20. He was referred to an oncology service for further treatment.

Table 1 shows a complete summary of all cases.

Discussion

HL is a type of malignancy that originates from B lymphocytes and spreads through and to lymph node groups or organs outside lymphatic system. In 1832, Thomas Hodgkin reported 6 cases of primary lymph node tumors [1]. He was the first to describe lymph system abnormalities in a paper titled “On some morbid appearances of the absorbent glands and spleen” [2]. In 1898 and 1902, Carl Sternberg and Dorothy Reed Mendenhall discovered giant cells in tissues of patients with HL, currently known as Reed-Sternberg cells [3]. Wolpaw et al. [4], Stolberg et al. [5], and Sheinmel et al. [3] subsequently described the radiology and pathology of HL characterized by Reed-Sternberg cells [5, 6]. Despite the descriptive nature of these papers, they provide a useful foundation to study HL. Subsequently, Macdonald [7] collected data from 284 patients diagnosed with HL in Nottingham from 1960 to 1975 and found that 43% had lung involvement. The most common radiographic type was peribronchial infiltration, which occurs in the early stage of the disease, whereas less common types, such as homogeneous or pneumonic infiltrates or nodules, usually occur later in the disease process. Radin [8] examined 60 cases of PPHL and reported that this disease was predominant in younger women. Between 1990 and 2003, 7 new cases of PPHL were described, and 5 additional cases were subsequently described in 2006 [9]. Specifically, a report from 1998 describes 3 cases of PPHL, 2 of which exhibited unilateral upper lobe cavitory lesions, and 1 exhibited bilateral lower lobe nodules [10]. Moreover, a study from India reported a thick-walled single cavitory lesion in the left upper zone with areas of necrosis observed on radio-

graphs [11]. Our experience and reports from Saudi Arabia indicate 2 reported cases of PPHL, and radiological studies showed pulmonary parenchymal masses [12].

Pulmonary involvement occurs in 15–40% of HL cases [13] and may be caused by peribronchiolar adenopathies near the pulmonary parenchyma or a significant amount of lymphoid follicles [14]. Pulmonary HL usually manifests as a single nodule, multiple nodules, a single cavitory lesion or a mass. However, we herein describe an unusual radiological feature – a multiloculated cavitory lesion. The differential diagnosis of cavitory lesions is complex and includes infectious causes, such as pulmonary Gram-negative bacteria, actinomycosis, histoplasmosis or aspergillosis, pneumonia, abscesses, pulmonary TB, hydrated cysts, or septicemic emboli. Moreover, these lesions may be due to a malignancy, such as squamous cell carcinoma, sarcomas, osteosarcomas or a metastasis to the lung. Nevertheless, this condition may also be attributed to granulomatous diseases, such as Wegener granulomatosis [13, 15, 16].

Because of this complexity, all patients underwent noninvasive and invasive testing. A septic workup excluded infectious disease, and this conclusion was supported by a lack of response to antibiotic therapy in cases 1 and 2. The chest CT scans of all patient showed enlarged reactive mediastinal lymph nodes, which may be attributed to the disease, a reaction to a parenchymal infection within the cavity or a pneumonic reaction. Considering the geographical origin of these cases, this presentation is usually due to an infective cause, but lymphoma should be considered as a cause, especially in young females.

Conclusion

Based on a literature review and our limited experience, we recommend that PPHL be included in the differential diagnosis of any cavitory pulmonary single or multiloculated lesion, especially in young females.

Statements of Ethics

The authors have no ethical conflicts to disclose.

Disclosure Statement

The authors have no conflicts of interest to declare.

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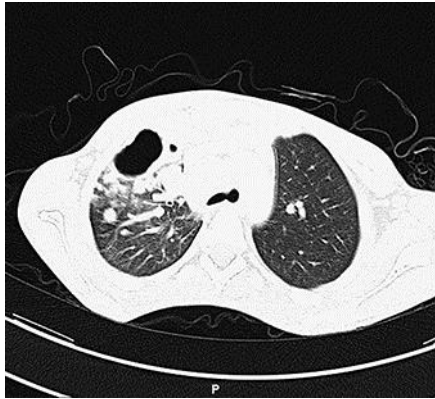


Fig. 1. Enhanced chest CT scan axial cut (pulmonary window) showing a large multiloculated cavity lesion involving the right middle lobe and the anterior mediastinum with internal air and fluid.

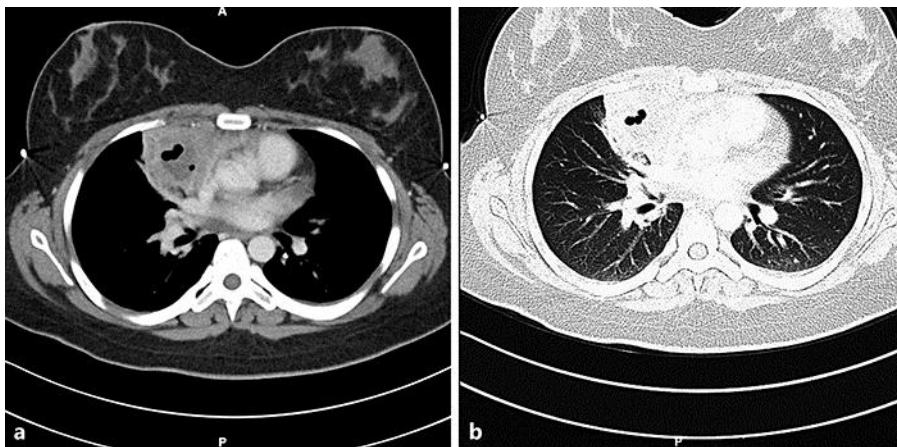


Fig. 2. a CT scan of the chest mediastinal window. b CT scan of the chest lung window.



Fig. 3. **a** CT scan of the mediastinal window. **b** CT scan of the lung window.

Table 1. Case summary

| | Patient 1 | Patient 2 | Patient 3 |
|--------------------------|---|---|---|
| Age, years | 16 | 29 | 21 |
| Gender | Male | Female | Female |
| Presenting complain | fever weight loss productive cough | productive cough hemoptysis | fever night sweat weight loss |
| Labs (abnormal results) | CBC: – WBC: 35,000/ μ L – Hgb: 8 g/L (13–18) – ESR: 120 mm/h (0–20) – CRP: 14 mg/dL (0–0.3) | CRP: 5.5 mg/dL ESR: 84 mm/h | CBC: – Hgb: 9.2 g/L – CRP: 5.8 mg/dL – ESR: 120 mm/h |
| Chest X-ray | large cavitory lesion with smooth margins and thickened walls; air fluid levels associated with adjacent lung consolidation involving the right middle and lower lung zones silhouetting the cardiac border | cavitory lesion involving the right mid zone of the lung | diffuse heterogeneous opacity occupying the right middle zones with evidence of a cavity |
| CT scan | large multiloculated cavitory lesion involving the right middle lobe and the anterior mediastinum with internal air and fluid; some reactive mediastinal lymph nodes were observed | 4.4×4.6×6.3 cm mass with central hypodensity indicating a multiloculated cavitory lesion; air was observed in the right anterior mediastinum and anterior segment of the right upper lobe; enlarged reactive lymph nodes were observed along the prevascular space and along the para-aortic region | large mass extending from the anterior aspect of the mediastinum with an intrapulmonary component measuring 11×9×12 cm, necrotic center and thick enhancing soft tissue rim indicating a multiloculated cavitory lesion |
| Biopsy source | mediastinal lymph node | mediastinal lymph node | cervical lymph node |
| Methods of biopsy taking | VATS mediastinoscopy | CT-guided biopsy | CT-guided biopsy |
| Histopathology | positive for CD30, CD68, CD3, CD20, and LCK | positive for CD30, CD15 | positive for CD30, CD15 |
| Diagnosis | nodular sclerosing type Hodgkin lymphoma | nodular sclerosing type Hodgkin lymphoma | nodular sclerosing type Hodgkin lymphoma |