

Case Report

Infectious giant bulla associated with lung cancer^{*}, ^{**}

Bolha gigante infecciosa associada a câncer de pulmão

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Abstract

A 79 year-old man sought treatment in the emergency room complaining of persistent fever, chest pain, and general fatigue. A chest X-ray showed a giant infectious bulla (24 cm in diameter) in the left lung. The patient had no history of abnormalities on X-rays, and his latest medical check-up, conducted in the preceding year, had produced no abnormal findings. Diagnostic procedures, including bronchoscopy, revealed lung cancer (large cell carcinoma) in the left lower bronchus. The tumor obstructed the airway. Although there have been various reports of giant bullae, their etiology remains unknown. We suggest that an obstruction, such as that caused by the tumor in this case, can lead to air trapping, resulting in the formation of a bulla. In the case of a giant bulla that rapidly increases in size, lung cancer should be included in the differential diagnosis.

Keywords: Lung neoplasms/complications; Infection; Drainage.

Resumo

Um homem de 79 anos procurou tratamento no pronto-socorro com queixas de febre persistente, dor torácica e fadiga geral. A radiografia de tórax mostrou uma bolha gigante infecciosa (24 cm de diâmetro) no pulmão esquerdo. O paciente não tinha histórico de anormalidades em radiografias, e seu último check-up médico no ano anterior não revelou anormalidades. Os procedimentos diagnósticos, incluindo broncoscopia, revelaram câncer de pulmão (carcinoma de pulmão de células grandes) no brônquio inferior esquerdo. O tumor obstruía a via aérea. Apesar de vários relatos de bolhas gigantes, a sua etiologia ainda é desconhecida. Nós sugerimos que uma obstrução, como a causada pelo tumor neste caso, pode causar aprisionamento aéreo, resultando na formação de uma bolha. No caso de uma bolha gigante que cresce rapidamente de tamanho, o câncer de pulmão deve ser incluído no diagnóstico diferencial.

Descritores: Neoplasias pulmonares/complicações; Infecção; Drenagem.

Introduction

Many aspects of giant bullae remain unknown. In addition, guidelines for the management of infectious giant bullae have yet to be established. In general, antibiotic therapy alone is insufficient to treat infectious giant bullae. Although surgical treatment is often recommended for patients with this condition, many such patients are not candidates for surgery because of poor general health status and high-risk factors. Here, we present a case of infectious giant bulla that rapidly increased in size.

Case report

A 79-year-old man sought treatment in the emergency room, complaining of a 3-month history of persistent fever, chest pain, and general fatigue. He had a smoking history of 60 pack-years and had been a heavy drinker of Japanese sake (180 mL/day) for the past thirty years. Laboratory test results indicated severe inflammation, and a chest X-ray showed a giant bulla with a diameter of 24 cm in the left lung (Figure 1a). By his own account, he had no history of abnormalities on X-rays, and his latest medical check-up, conducted in the preceding year, had

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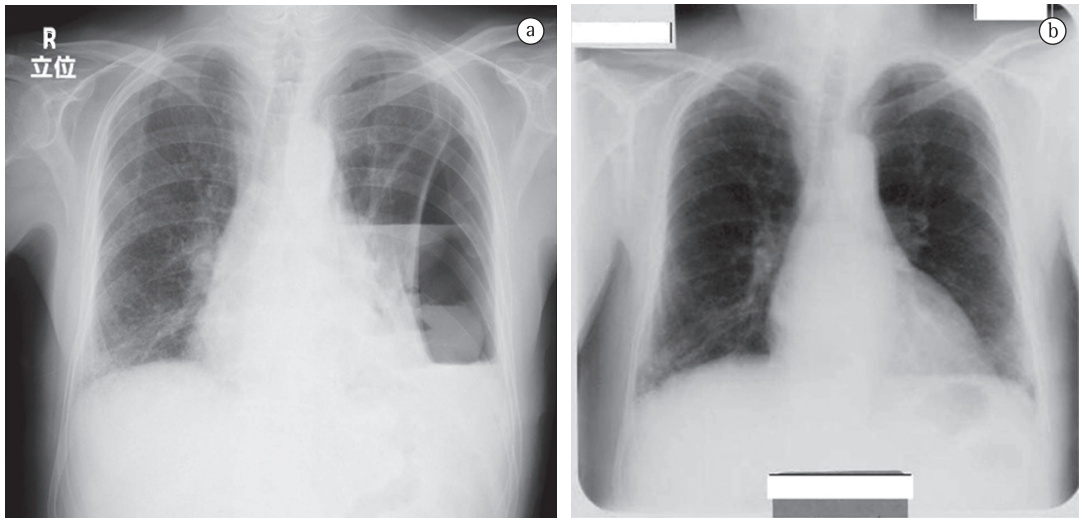


Figure 1 – Chest X-rays: (a) at admission, showing a giant bulla with an air-fluid level in the left lung (left); (b) at one year prior, showing no abnormalities.

produced no abnormal findings (Figure 1b). CT scans of the chest revealed that the bulla was fluid-filled and measured $4.5 \times 17 \times 24$ cm (Figure 2). The patient was diagnosed as having a lung abscess and was admitted to our hospital.

Sputum and blood cultures were obtained. Parenteral antibiotic therapy was started with the tazobactam-piperacillin combination (13.5 g/day). Bronchoscopy on post-admission day 14 showed an obstruction in the left lower bronchus (Figure 3), and a biopsy was taken. Pathological examination of the biopsy specimen revealed large cell carcinoma. Using the tumor-

node-metastasis (TNM) staging system, we determined the clinical stage of the tumor to be T2N2M1 (stage IV), because there were multiple metastases to the left adrenal gland and third lumbar vertebra, as revealed by (18)F-fluoro-2-deoxyglucose positron emission tomography.

The cultures showed no growth, and the antibiotic therapy was maintained, despite a number of changes to the drug regimen. Finally, a percutaneous drain was placed into the infectious bulla. After the bulla had been drained, the symptoms and laboratory test results improved temporarily. However, on post-

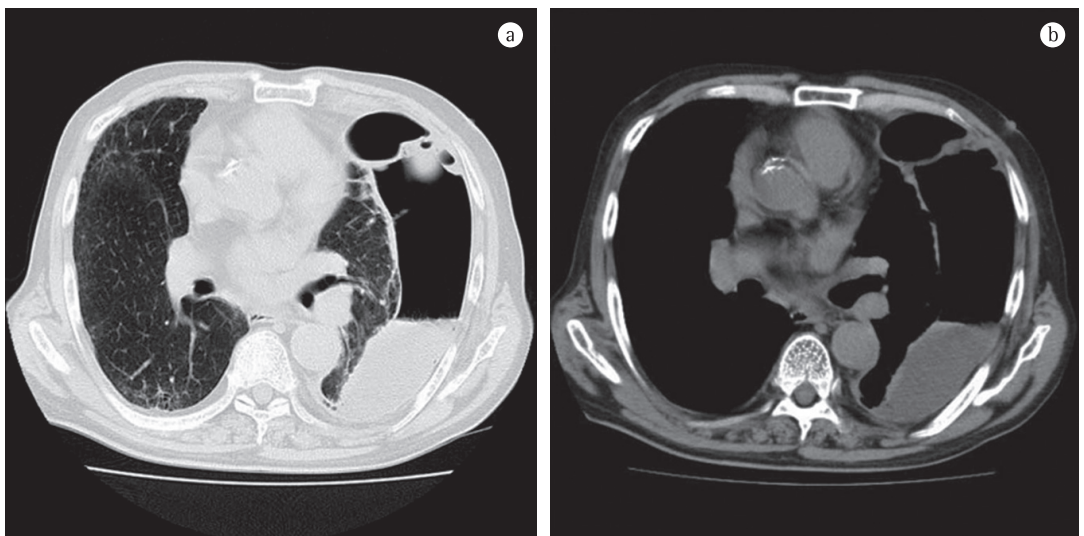


Figure 2 – Chest CT at admission, lung and mediastinal window settings (a and b) showing a fluid-filled bulla, measuring $4.5 \times 17 \times 24$ cm, in the left lung.

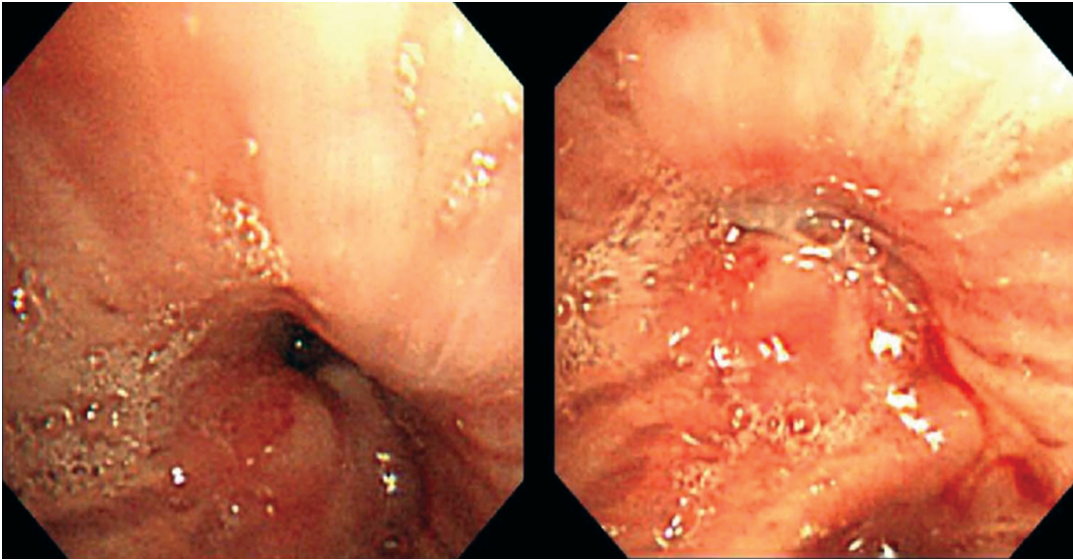


Figure 3 – Bronchoscopic view of an obstruction (tumor) in the left lower bronchus.

admission day 34, the patient died from fungal sepsis and multiple organ failure. No autopsy was performed, because the family did not consent.

Discussion

There is no established definition of a giant bulla, the various definitions including a bulla that occupies at least one third of a hemithorax,⁽¹⁾ one that occupies at least one half of a hemithorax,⁽²⁾ and one that is larger than 10 cm in diameter.⁽³⁾ Giant bullae do not participate in gas exchange, and, because they occupy a considerable amount of space, they can impair respiratory mechanics, thereby increasing the work of breathing, reducing exercise capacity, and causing dyspnea.⁽¹⁾ In particular, bullae in the middle or lower lobes severely affect the diaphragm, impairing pulmonary function.^(4,5) Nomori et al.⁽⁶⁾ reported that patients undergoing video-assisted thoracoscopic surgery showed significant postoperative improvement in pulmonary function parameters such as FVC and FEV₁. In the case presented here, the patient showed an improvement in SpO₂ (from 94% to 98%) after percutaneous drainage.

Although various theories regarding the etiology of giant bullae have been proposed, the origin of such bullae remains controversial. It has been suggested that their occurrence is due to the so-called “check-valve mechanism”,⁽³⁾ bronchial occlusion, or transformation of the

pulmonary and bronchial arteries.⁽⁷⁾ In fact, the only other mammal in which pulmonary emphysema spontaneously occurs is the horse, and an experimental study in horses indicated that bronchial artery occlusion plays an important role.⁽²⁾ Most investigators agree that degenerative lesions cause the alveolar walls to lose their elasticity.⁽²⁾ As the walls expand, the alveoli enlarge and some rupture, allowing air to escape into the lung parenchyma, resulting in the formation of blebs and bullae.⁽²⁾ Fain et al. attributed the formation of giant bullae to smoking, which decreases the blood flow to the alveoli.⁽²⁾ In the case presented here, the giant bulla appeared to have grown rapidly, given that no abnormalities had been found during the medical check-up conducted in the preceding year. We presume that the check-valve mechanism related to the tumor in the left lower bronchus promoted the growth of the bulla.

It has not been established whether giant bullae are more likely to occur on one side or the other. Although many authors have reported that the right side is the more common location,^(5,7) others insist that no such sidedness exists.⁽⁸⁾ It appears certain that the majority of patients with giant bullae are male and are (current or former) heavy smokers.^(5,7-9) It was recently reported that giant bullae are associated with lung cancer.^(5,9-12) Nakamura et al. demonstrated that lung cancer can arise from the wall of a giant bulla and therefore recommended that such bullae always be resected.⁽⁸⁾ The most common clinical

symptom in patients with giant bulla is chest pain. Consequently, giant bullae are commonly misdiagnosed as pneumothorax, as occurred in the case present here.

As previously mentioned, infectious giant bullae cannot be successfully treated through antibiotic therapy alone. Many physicians believe that cases of infectious giant bullae require surgical treatment. The most widely used surgical procedure for the resection of giant bullae is the method first described in 1947 by Naclerio & Langer.⁽¹³⁾ Patients who are not candidates for surgery have been successfully treated with various less invasive treatments, such as bronchial embolization by bronchoscopy,⁽¹⁴⁾ percutaneous drainage using a pigtail catheter,⁽¹⁵⁾ combined bronchial embolization and percutaneous drainage,⁽¹⁶⁾ and repeated pleurodesis.⁽¹⁷⁾ Wali et al. reported a 9.7% rate of complications related to percutaneous drainage, the most common complications being catheter occlusion, chest pain, pneumothorax, and hemothorax.⁽¹⁵⁾ Studies have shown that pneumothorax occurs in 19–60% of patients undergoing CT-guided lung biopsy,^(18–20) the complication rate related to percutaneous drainage being therefore acceptable by comparison.

In conclusion, when a patient with infectious giant bulla is not a candidate for surgery, percutaneous thoracic drainage should be considered as an early intervention, because it can prevent the worsening of the general health status of the patient. In the case of a giant bulla that rapidly increases in size, lung cancer should be considered in the differential diagnosis.

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