Spontaneous Pneumothorax as a Complication of Cystic Bronchioloalveolar Carcinoma: Pathology, HRCT Findings and Review of the Literature

Komplikasyon Olarak Spontan Pnömotoraks Gelişen Kistik Bronşioloalveolar Karsinoma Olgusu: Patoloji, YRBT Bulguları ve Literatürün Derlenmesi

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ABSTRACT

We present a rare case of cystic bronchioloalveolar cacinoma (BAC) which was complicated by spontaneous pneumothorax. Descriptions about the mechanism of cysts and complications of pneumothorax in BAC are few in the literature. Check-valve mechanism is one of these factors which may be responsible for the cystic lesions and spontaneous pneumothorax in cystic BAC. Narrowing of the respiratory bronchioles by the tumor cells should be a cause of the check-valve mechanism. In our case, high resolution CT (HRCT) showed that some of the cysts have a connection with the airways, which was confirmed by pathology. This case is original because the connection between the airways and cysts was supported by not only pathology specimens but also HRCT. *(Turk Toraks Derg 2012; 13: 178-80)*

Key words: Bronchioloalveolar, carcinoma, spontaneous, pneumothorax, computed tomography

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ÖZET

Komplikasyon olarak spontan pnömotoraks gelişen kistik bronşioloalveolar karsinoma (BAK) olgusunu sunuyoruz. Kistik BAK'da kistlerin ve pnömotoraks oluşumunun olası mekanizmalarından literatürde azda olsa bahsedilmiştir. Kistik BAK'de kistlerden ve spontan pnömotorakstan sorumlu tutulan mekanizmalardan biriside engelleyen kapak mekanizmasıdır. Tümör hücrelerinin respiratuvar bronşiollerde daralmaya neden olması bu engelleyen kapak mekanizmasının nedenlerinden birisi olabilir. Bizim vakamızda da bu kistlerin küçük hava yolları ile olan bağlantısı yüksek rezolüsyonlu bilgisayarlı tomografi (YRBT) tetkikinde gösterilmiş ve bulgular patolojik olarak da kanıtlanmıştır. Kistik BAK'ın komplikasyonu olarak spontan pnömotoraks gelişen bu olgunun özgünlüğü olguda kistlerin havayolları ile olan bağlantısının sadece patolojik olarak değil aynı zamanda YRBT ile de gösterilebilmesidir. *(Turk Toraks Derg 2012; 13: 178-80)*

Anahtar sözcükler: Bronşioloalveolar, karsinoma, spontan, pnömotoraks, bilgisayarlı tomografi

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INTRODUCTION

Bronchioloalveolar carcinoma (BAC) is a polymorphic lung cancer which has an increasing incidence. The range of BAC in all pulmonary lung cancer is 2-10% [1]. Most common types are a solitary nodule, multiple nodes and multiple areas of consolidation. Cavitations are uncommon for BAC but they can present as single or multiple lesions [2]. Isolated cases of multiple cavitary lesions in BAC have been reported [3]. Some cases with the pathological diagnosis of BAC which were detected before 1999 are reclassified as adenocarcinomas after the recent reclassification of lung and pleural tumors. Although the cavitation or cystic change is reported in 7% of all BAC, most of these are generally suggestive of tumor necrosis and invasive adenocarcinoma [4]. According to recent reclassification, the ratio of the cystic BAC is likely to be rarer than 7%. Descriptions about the mechanism of cysts and complications of pneumothorax in BAC cases are few. We present a rare case of cystic BAC with radiological and pathological images.

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CASE REPORT

A-76-year old man was admitted to our hospital with left chest pain and shortness of breath for 3 days. The physical examination finding at hospital admission was decrease of breath sounds in the left hemithorax. There were no rales or ronchus at auscultation. Body temperature and complete blood count were within normal range. Arterial oxygen saturation was 97%. The patient had been a smoker for 30 years (1 package per day). There was no history of trauma. Posteroanterior chest radiograph showed a pneumothorax on the left side and diffuse acinary opacities in the bilateral lower regions of the lung (Figure 1). A chest tube was inserted into the left side for the pneumothorax. High-Resolution Computed Tomography (HRCT) showed bilateral, multiple, thick and thin walled cystic lesions. Pulmonary acinary nodules and consolidation areas were also present (Figure 2). Some of the cystic lesions were located in the subpleural spaces and peripheral area of the lung (Figure 3). Some of these cysts had a connection with the airways. Ground-glass appearance and consolidation around the cysts with bronchiectasis were also present (Figure 4). After the radiological findings bronchoscopywas carried out in the patient. There were normal findings at bronchoscopy. The cytology of the bronchioloalveolar lavage was normal. The histological, cytological and bacteriological examination of the transbronchial biopsy specimen were also normal. Open left lower lobe biopsy was performed. The pathological diagnosis was BAC (Figure 5). Radiotherapy and chemotherapy were not suitable for the patient because of the hypoxemia, extensive tumoral infiltration of lung and failure of general health condition. Oxygen concentrator and

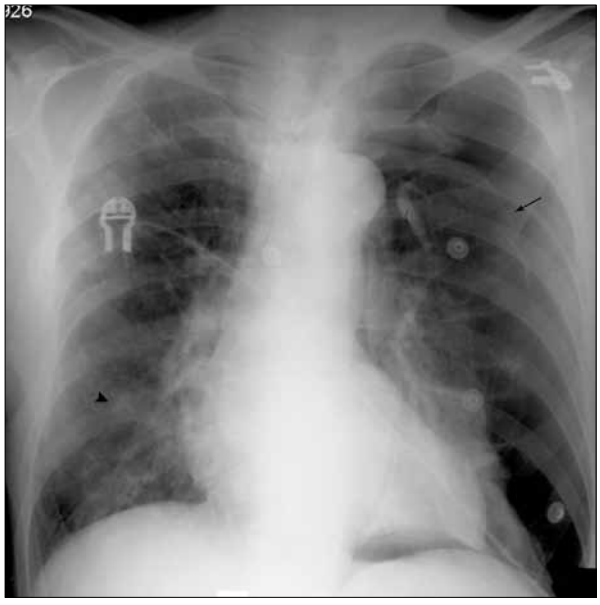

Figure 1. X-ray chest radiography at admission to hospital, demonstrating pneumothorax on the left side [black long arrow] and aciner opacities in the right paracardiac spaces [black arrow head]

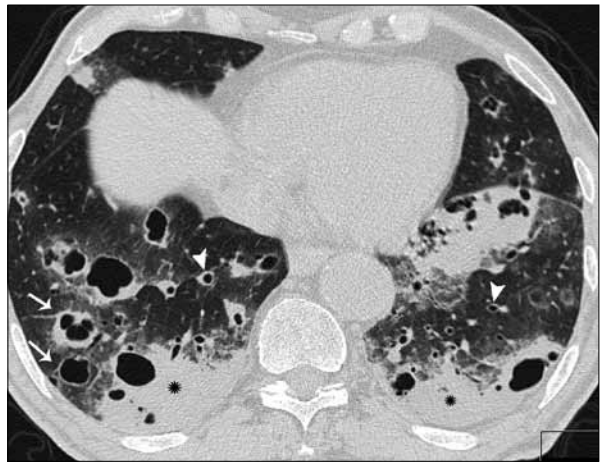

Figure 2. HRCT scan shows thin and thic walled cysts [white arrows], bronchiectasis [white arrow head], consolidation and ground glass appearance [black asterix] at the base of lung

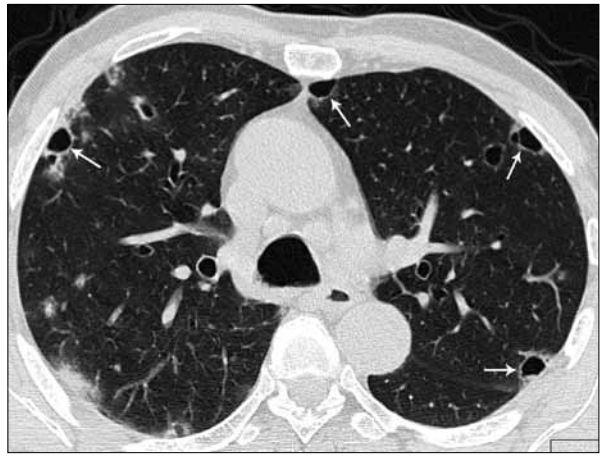

Figure 3. HRCT scan shows the cystic lesions in subpleural spaces [white arrows]

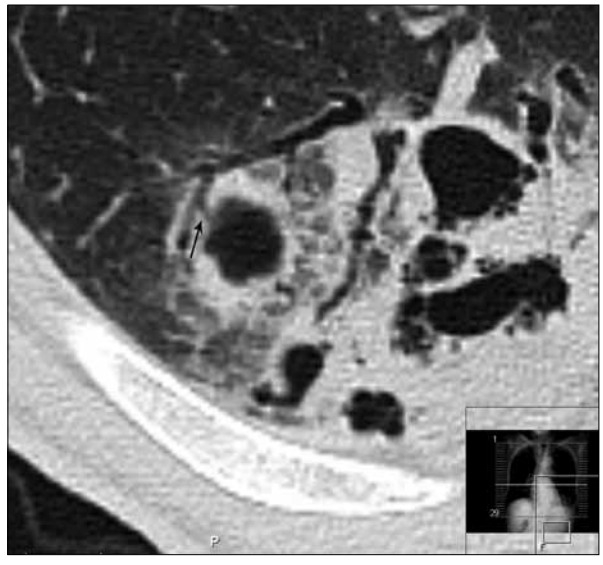

Figure 4. The interaction between the thick walled cyst and the ectatic terminal bronchiole [black arrow] can easily be seen in this magnified HRCT scan. Also note the cysts and bronschiectasis in the consolidation area

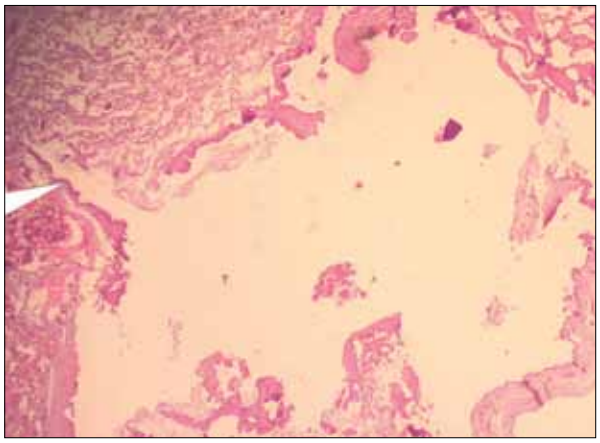

Figure 5. A photomicrograph of a section of lung obtained at open lung biopsy, demonstrating narrowed terminal bronchiole lined by columnar cancer cells [white arrow head] with lepidic growth of broncioloalveolar cell carcinoma [hematoxylin-eosin, original magnification x 100]

steroid treatment were planned to alter the hypoxemia. The patient was discharged and called for routine clinical follow up.

DISCUSSION

The cystic lesions in BAC are well described and various causes have been proposed [1,3,4]. Spontaneous pneumothorax is also described as a complication of lung cancer [5]. Also, the probable mechanism in relation to histological findings has been discussed [3]. Weisbord et al. indentified thin-walled cystic lesions in 4 patients with BAC. The authors suggest 3 mechanisms to explain formation of cystic lesions. (a) ischemic necrosis (according to the revised WHO classification this pattern is reclassified as adenocarcinoma). (b) obstructive bronchiectasis by check-valve bronchiolar obstruction (The check-valve mechanism was thought to be the cause of cystic lesions, however, the explanation for the mechanism was increased thickness of the terminal bronchioles due to fibrosis). (c) air containing neoplastic space [4]. Morimoto et al. [6] suggested that the mechanism of cystic change in BAC is disruption of the alveolar structure and enlargement of disrupted spaces or the check-valve mechanism. However, they could not document the mechanism of cystic change clearly. Kobayashi et al. [7] suggested that the majority of the cavities in BAC were characteristic tension cavities or cysts resulting from an increased intraalveoler pressure caused by mucus produced by the tumor cells in the alveolar space. Air trapping and hyperinflation of the distal lungs occur when the terminal bronchioli are narrowed or occluded, such as in bronchiolitis [8]. Minami et al. [9] also underlined the check-valve mechanism as a reason for cysts in BAC. However they did not present either HRCT or CT images of the case. They had proven this mechanism with only autopsy findings. Small airways are narrowed when they are invaded with tumor cells, such as BAC. This can make a check-valve mechanism which causes the cystic dilatation of the secondary pulmonary lobules. The air trapment in the distal lobuler bronchioli makes the tension rise in the cyst and it eventually ruptures. In BAC, the peripheral bronchi are narrowed by malignant invasion and multiple cavitary lesions form due to check-valve mechanism [3,10]. This mechanism may be the cause of spontaneous pneumothorax also. Excitingly, our patient's HRCT showed some of the cysts were connected with the distal airways, which was also confirmed by pathology. the pathology specimen showed narrowed distal airways opening to the cysts and distended alveolar spaces. Another interesting finding in HRCT was the peripherally located cysts resembling the paraseptal emphysema, which could be the cause of spontaneous pneumothorax. To our knowledge, this is the first case of cystic BAC with spontaneous pneumothorax which represents the check-valve mechanism proved with not only pathology specimens but also HRCT.

In summary, cystic BAC complicated by spontaneus pneumothorax is a very rare entity and HRCT should be kept in mind to demonstrate the connection of the cavities with distal airways in similar cases.

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