

Pulmonary hamartoma mimicking lung tuberculoma in tuberculosis patient – a case report

Hamartoma płuc diagnozowana jako gruźliczak u chorego z gruźlicą płuc – opis przypadku

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Abstract

Pulmonary hamartomas are benign tumors, which represent abnormal mixture of fibrous connective tissue, hyaline cartilage, fat and bone as well as bronchial epithelial cells. Differential diagnosis of tuberculoma, malignant neoplasms and hamartoma is necessary. We present the tuberculosis patient with pulmonary hamartoma mimicking lung tuberculoma. The uncorrected diagnosis of tuberculoma was made based on symptoms and CT imaging. However, histopathologic analysis of surgically enucleated tumor confirmed non-infectious origin of the lesion (hamartoma with hyaline cartilage predominance).

Key words: pulmonary hamartoma, tuberculoma, tuberculosis.

Streszczenie

Hamartoma płuc jest niezłośliwym guzem zbudowanym z chaotycznie ułożonej mieszaniny tkanki łącznej, chrząstki szklistej, tkanki tłuszczowej, kości i komórek nabłonka oskrzeli. Hamartomę należy różnicować z gruźliczakiem i rakiem płuca. Opis przypadku dotyczy chorego z gruźlicą płuc i współistniejącą hamartomą płuc zdiagnozowaną błędnie jako gruźliczak na podstawie objawów i obrazu tomografii komputerowej. Badanie histopatologiczne usuniętego operacyjnie guza potwierdziło diagnozę hamartoma o przeważającym utkaniu chrząstki szklistej.

Słowa kluczowe: hamartoma płuc, gruźliczak, gruźlica.

Introduction

Hamartomas are tumours of benign character, which represent abnormal mixture of mature tissue chaotically located within the organ of hamartomas' origin. Hamartomas are presented as solitary pulmonary nodule (SPN) described as well-circumscribed, rounded, smooth edged, dense (without cavitation) pulmonary lesion, 3 cm or less in diameter. Pulmonary hamartomas are the third most common cause of SPN (5%-8%), just after malignant neoplasms and granuloma formations caused by *Mycobacterium tuberculosis*, atypical mycobacteria or fungal infections [1]. Most hamartomas are discovered incidentally and commonly among man, with the peak incidence during sixth or seventh decade of life [2]. In a large autopsy series from Mayo Clinic, pulmonary hamartomas were found in 2 of 7972 cases (0.025%) but incidence of pulmonary hamartomas may reach 0.3% of general population [3].

Case report

A 54-year-old male patient presented with a round, smooth, well-defined, 2.5 cm in diameter lesion, with calcification, located on the apex of the left lung in clavicle "shadow" (Fig. 1A). In the initial diagnosis, the X-ray in oblique projection of lung apex was done for the differentiation from clavicle osteosclerosis (Fig. 1B). Hemoptysis was an individual symptom of the disease. Positive tuberculin test was 30 mm in diameter but *Mycobacterium tuberculosis* was not detected in microscopic slides as well as in standard culture. Bronchofiberscopy examination and histopathologic examination of material from intrabronchial biopsy revealed chronic bronchitis. Family history of tuberculosis occurrence was negative but the tuberculosis was diagnosed and successfully treated when patient was 5 years old.

A computed tomography scan of left lung apex showed well-limited, irregular tumor (26 × 23 mm) with characteristic

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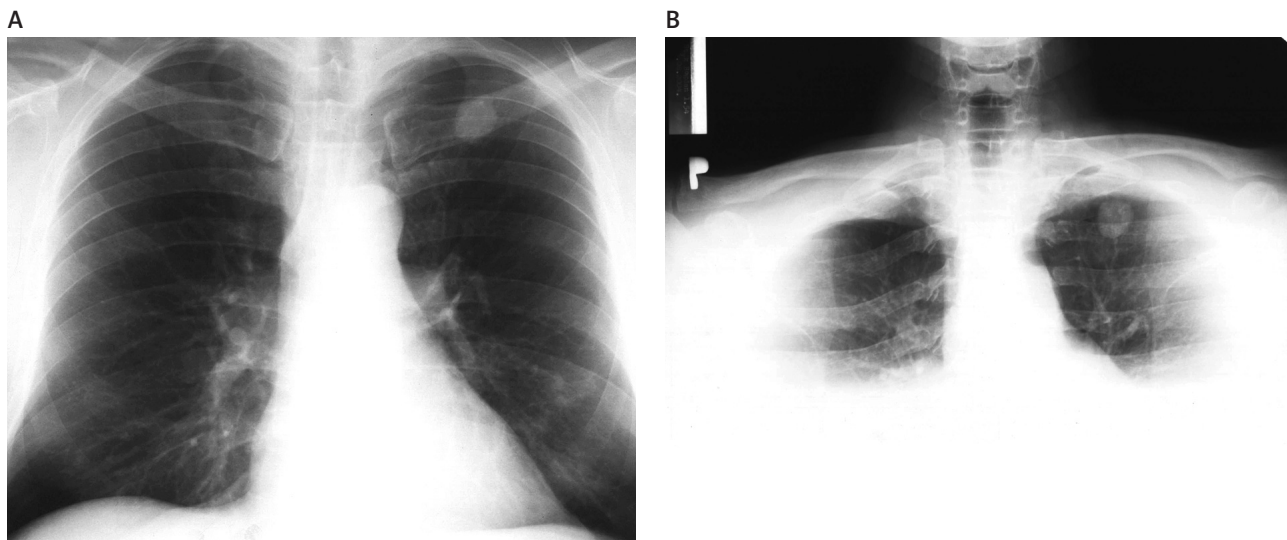


Fig. 1A–B. Chest X-ray examination showed round, smooth, well-defined lesion in anterior-posterior (A) and oblique (B) projection

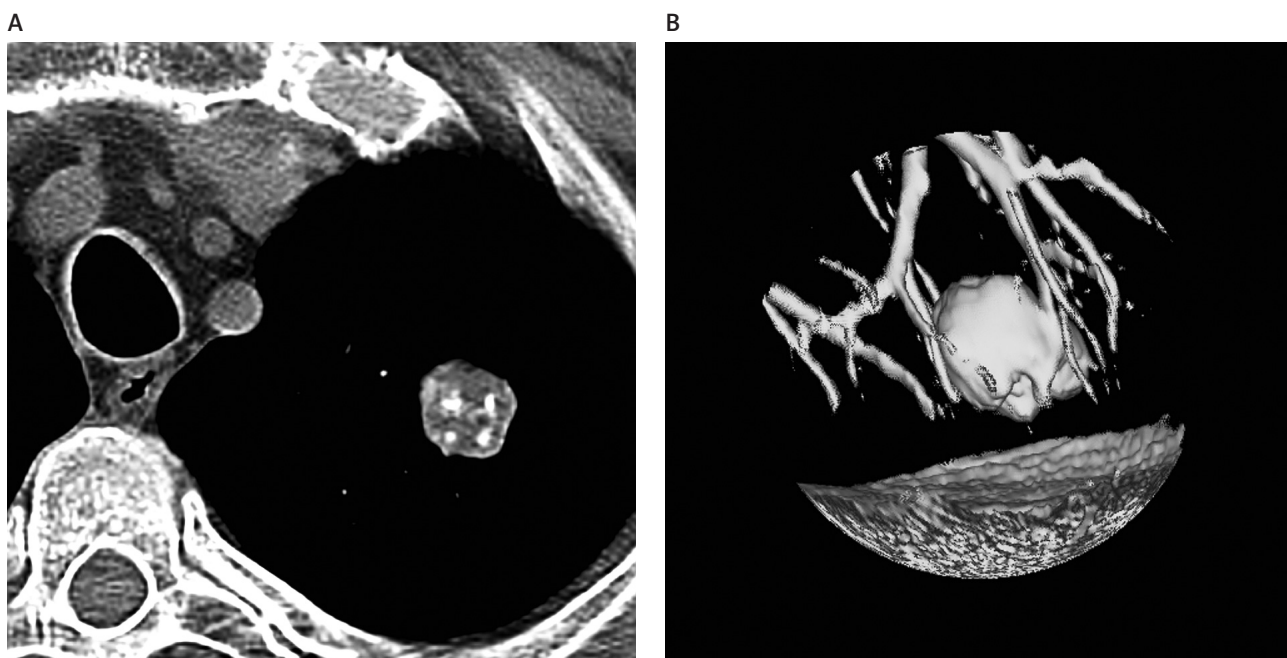


Fig. 2A–B. Computed tomography showed irregular tumor with characteristic popcorn pattern of calcification (A) and 3D visualization of tumor and vessels (B)

popcorn pattern of calcification (Fig. 2A, 2B). Density of the lesion ranged between -1 UH (small area) to 1354 UH (calcification). Inferior lobe of the left lung showed reduced volume with the presence of calcified tumor covered by fibrosis (14×11 mm) (Fig. 3A). This change corresponded to non-active tuberculosis. Moreover, several micronodular, intralobular changes encircled by ground glass opacities and expressing “the tree-in-bud” sign were found in inferior lobe of left lung (Fig. 3B). These lesions have been interpreted as bronchogenic tuberculosis dissemination.

Based on symptoms and CT imaging, tuberculosis and suspicion of tuberculoma was diagnosed. Patient started treatment with standard dose of rifampicin, isoniazid and pyrazinamide. In the antituberculous drug cover, the patient

was qualified to surgical resection of lesion. The tumor was enucleated. On pathologic examination the resected lesion measured 20×15 mm and a cut surface revealed gray and compact tissue. Surprisingly, the microscopic examination showed tumor which composed of mature hyaline cartilage. Based on these findings, pulmonary hamartoma was diagnosed and antituberculous treatment was continued what resulted in disappearance of changes visible on X-ray.

Comment

90% of pulmonary hamartomas are located peripherally with only occasional lesions being central or endobronchial in location [4]. Parenchymal hamartoma consists of fibrous

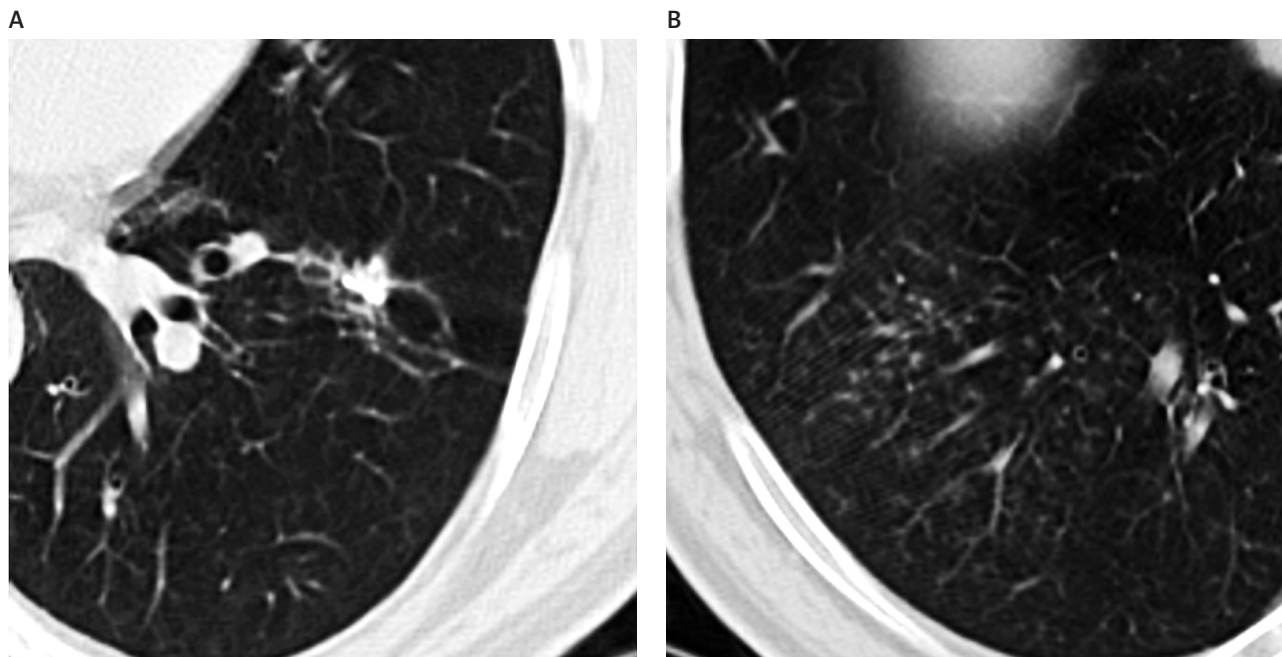


Fig. 3A–B. The changes corresponded to non-active tuberculosis (calcified tumor covered by fibrosis – **A**) and bronchogenic tuberculosis dissemination (**B**)

connective tissue, hyaline cartilage, fat and bone as well as bronchial epithelial cells arranged in disorganized fashion. Hamartomas are rarely symptomatic but in some cases are associated with hemoptysis and cough. The characteristic radiographic features of hamartoma are presence of popcorn-like calcification (seen in 10-15% of patients) but this is not diagnostic since calcifications may appear in tuberculosis as well. Therefore, differential diagnosis of tuberculosis and hamartoma is necessary, what our case report confirms [3, 5, 6]. Histopathological analysis of material from transthoracic needle aspiration biopsy (less than 85% of cases), transbronchial biopsy or surgical resection confirmed diagnosis of hamartoma [6]. Moreover, pulmonary hamartoma in our patient was predominantly cartilaginous, which only occurs in 1% of hamartomas [7]. Interestingly, there are reports that several pulmonary chondroid hamartomas had chromosome rearrangements involving the chromosomal regions 12p15 and 6p21. These regions include *HMG1-C* and *HMG1(Y)* genes, that encode DNA-binding proteins with both chromatin structural and gene regulatory roles [8, 9].

Tuberculoma is an uncommon manifestation of tuberculosis. Similarly to hamartoma, X-ray examination typically showed a well-marginated round or sometimes oval opacity typically in the upper lobes. As necrosis and chronic disease occurs, calcification may be observed in the centre of tuberculomas. Generally the calcification is central but occasionally may involve most of the visible lesion. These lesions are generally stable for long periods of time demonstrating only slow growth over a period of several months or years. Cavitation is extremely rare within tuberculomas [10].

The relation between origin of tumor in lung apex and development of tuberculosis seems to be obvious, due to

diagnosis of tuberculosis in our patient. In our study, we presented for the first time the pulmonary hamartoma coexisting with tuberculosis.

In the majority of hamartoma patients, the tumor is removed by wedge resection or enucleation and patients require no further treatment. However, in our patient further antituberculous treatment was necessary.

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