Pulmonary hamartoma - case report and review of literature

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SUMMARY
Pulmonary hamartoma is the most common benign tumor of the lung. We report the case of a healthy, 46-year-old woman with an incidentally discovered chondromatous hamartoma, which has been excised thorascopically. She developed hamartoma within a year.

Key word: Hamartoma; Lung Neoplasms

INTRODUCTION
Pulmonary hamartomas (PH) also known as mesenchymomas, can be parenchymal (80%) or endobronchial (10–20%). Parenchymal lesions are usually an incidental finding and range in size from 1 cm to 8 cm in various series. The endobronchial tumors usually present with new onset of respiratory symptoms, which are most commonly recurrent chest infections or hemoptysis. Pulmonary hamartoma are the most common form of benign lung tumors with an incidence of between 0.025% – 0.32% according to different necropsy studies (1-3). We report a case of accidentally found pulmonary hamartoma in previously healthy woman who was admitted to emergency room because of strong pain in upper back on the day of admission.

CASE REPORT
A 46-years-old female, a worker by occupation and a chronic smoker, presented in emergency room with a strong pain in upper back. She felt it suddenly in working place. On admission, physical examination and laboratory investigations were normal. Per anamnesis, obtained data indicated a history of chronic gastritis. She was an average built, denied any significant loss in weight and fever. Plain chest radiograph showed a solitary nodular opacity of 1.8x1.8 cm in diameter (Figure 1).

Because of respiratory infection a year before, patient had chest X ray and the finding was normal. To find out the origin of pulmonary change the patient was examined by abdominal ultrasonography, computed tomography of chest and abdomen, gastroscopy, colonoscopy, breast ultrasonography, bronchoscopy, and gynecology. During clinical examination, pain in upper back disappeared after application of NSAIID. Gastroscopy confirmed the presence of chronic gastritis. Breast ultrasonography revealed hypoechogenic small nodule in left upper quadrant, 8mm in diameter. The results of mammography excluded of any suspected neoplasia. In the sample obtained by bronchoaspiration, we found staphylococcus coagulase negative species sensitive to cephalosporin, penicillin, and tetracycline. The evaluation of cytological analyses showed moderate population of squamous cell layered epithelium, mostly single and without cell nuclear anisotropy, but still present in smaller individual small groups with enlarged nuclei, and mild to moderate anisonucleosis- the finding suspected of malignancy. Patient was sent to the thoracic surgery for further diagnostic procedures. Thorascopic excision of the sample contained islands of cartilage, fat, fibromyxoid stroma, and narrow spaces lined by respiratory epithelium. Histopathological finding of the specimen confirmed the presence of chondroid hamartoma. After the procedure, patient was feeling well. She was advised for regular checkups.

DISCUSSION
Pulmonary hamartoma was first described by Albrecht in 1904. Pulmonary hamartoma was thought to arise from embryologic rests that were present in fetal life but generally did not become visible until adulthood. However, a cytogenetic analyses of the pulmonary hamartomas show an abnormal karyotype and reveal recombinations between chromosomal bands 6p21 and 14q24 (4), thus supporting the opinion that a hamartoma of the lung is a true neoplasm. However, the nosogenesis of PH remains unclear. Pulmonary hamartomas are usually found in adults with a peak incidence in the sixth decade. There is a male preponderance, the male:female ratio being 2:1 to 3:1. Pulmonary hamartomas are often asymptomatic and they are typically discovered as an incidental coin lesion on a routine chest radiograph. Radiologically, hamartomas account for 7% to 14% of pulmonary coin lesions (5). PH can occur in all parts of lung, but most often, they are found in the periphery and rarely near the hilar parts. On chest radiographs, pulmonary hamartomas characteristically appear as well-defined, solitary pulmonary nodules. They may show varying patterns of calcification, including an irregular popcorn, stippled, or curvilinear pattern, or even a combination of all 3 patterns. When calcification or fat is detected in a well-circumscribed peripheral lung tumor, a diagnosis of hamartoma can confidently be made. However, the characteristic calcification is seen in only approximately 15% of patients. Calcification in a pulmonary nodule on imaging indicates a high probability that the lesion is benign. However, not all calcified pulmonary nodules are benign and the differential diagnosis includes a primary central lung carcinoid, metastasis, and a primary bronchogenic carcinoma (6). Serial chest PHs radiographs may demonstrate slow growth. Rapid growth has rarely been reported. This feature may make the differentiation of a hamartoma from a bronchogenic carcinoma difficult (7).

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Today, even with the advancement in medical therapy, pulmonary resection remains the most important treat measure of patients with pulmonary hamartoma. Controversy, however, still exists about the indication and timing of surgery (8). Since most pulmonary hamartomas are nonexpanding or slowly growing neoplasms, some authors believe that surgery is necessary only when expansion is recorded in young or middle aged patients or accompanying obvious pulmonary symptoms. According to Gui and associates (9), the pulmonary hamartoma has the tendency of expansion or recurrence, and chronic inflammatory stimulation of local position may contribute to the development of malignancy. Therefore, when a solitary pulmonary lesion is more than 2.5 cm or the possibility of malignancy cannot be excluded, surgical resection is deserved to be performed and should be mandatory.

Our opinion is that pulmonary hamartoma is indicated for surgery in case of: (1) a solitary pulmonary lesion with diameter above 2.5 cm; (2) it is an overweight psychic burden for a patient; (3) having the tendency of expansion or recurrence; (4) pulmonary symptoms unresponsive to drug treatment; and (5) the lesion cannot be differentiated from malignancy.

In case of our patient initial diagnosis of metastatic disease was indicated by radiological finding of solitary lung nodule, which rapidly developed only one year after the normal X-ray finding. As bronchoaspiration findings referred to malignancy, surgical treatment was the only solution to establish diagnose. The case of our patient implies that hamartoma can be expressed in very short time, within a year.

Conflict of interest
We declare no conflicts of interest.

REFERENCES